The Role of Social Support in Helping People with EDS Manage Their Condition

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Declaration

This work is original and has not been submitted in relation to any other degree or qualification

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Department of Psychology

Research Module Meeting Log

NAME: Kate Appleby

SUPERVISOR: Dr. Janine Carroll

Date:	**Discussion topics**
22-01-16:	Potential topics for study; recommended reading
26-02-16:	Research question; interview questions; recruitment; location of interviews
11-03-16:	Ethics deadline (16-03-16); details of Ethics application; permission requirements for off-site location for interview
11-05-16:	Recruitment via EDS charity - their requirements; Ethics amendment forms: amendment to consent form required by Ethics committee; and video call interviews. Evidence of permission granted by online support group administrators to recruit participants
15-06-06:	Ethic amendment – additional interview location
06-07-16:	Progress update re interviews, and literature review
25-08-16:	(telephone) Progress update re introduction and literature review
02-09-16:	(telephone) Feedback on method
19-09-16:	(telephone) Themes
03-10-16:	(telephone) Introduction and Method
13-12-16:	Revised timetable
22-05-17:	(telephone) Progress update
05-06-17:	(telephone) Analysis

12-06-17: Themes discussion

26-06-17: (telephone) Potential themes revision

03-07-17: (telephone) Feedback on revised themes, discussion plan, and guidance on submission of dissertation

24-07-17: (telephone) Feedback on discussion and queries re final details – theme numbering, document format, supervisory log, and references

SIGNED

STUDENT _____ DATE: _____

SUPERVISOR _____ DATE: _____

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Abstract

The Ehlers-Danlos syndromes (EDS) are complicated, multisystemic disorders that can impact almost every aspect of an individual's life and severely affect functioning. Research into optimal long-term management of EDS is still sparse and recommended medical treatments are largely based upon the clinical opinions of healthcare providers. Social support is linked to coping with adversities such as chronic health conditions, and has a relationship with health-related quality of life. This qualitative study examined the role of support in optimal management of EDS. Five participants with EDS were interviewed, and thematic analysis was carried out. Four themes relating to disparate sources of support emerged: community incorporated personal or face-to-face support (e.g. spouse, family, friends, and peers). Medical support included specific professionals and types of care available to people with EDS. The role of self-support included and focused on resilience and how support helps development of resilience. The results of this study suggest that external support can facilitate the development of resilient behaviours and attitudes in individuals with EDS, which enable better management of their condition. Interventions that incorporate appropriate, multidisciplinary, medical care with the goals of improving individuals' access to support, such as information and peer support, could enable better management of EDS. Optimal management of EDS is likely to result in better HrQoL and reduce healthcare-related expenditure associated with treatment of EDS.

Introduction

Long term conditions (LTC), or chronic illnesses, can be highly disabling and often require care from healthcare professionals. LTC include diseases such as diabetes, dementia, arthritis or heart disease, and people with LTC often require support in order to effectively manage their condition(s). An increasing proportion of national healthcare expenditure is spent on LTC. It has been reported that 70% of the total health and care expenditure in England was associated with the treatment and care of the 30% of the population with one or more LTC (House of Commons Health Select Committee, 2014). Further, the number of people with one or more LTC (at the time of the report; 15 million) was expected to increase to approximately 18 million by 2025, and it was predicted that healthcare costs associated with the treatment and management of LTC would rise by around £5 billion annually between 2011 and 2018. As such, effective management of LTC is a major public health issue with serious implications for the allocation of national healthcare resources.

LTC necessitate complex care plans that may require input and management by multiple healthcare professionals, potentially from different healthcare specialisations, and this can prove costly. A number of LTC cannot be cured; however effective long-term management can improve an affected person’s quality of life (QoL), and in some cases life expectancy (Thomas, 2009). The LTC that have the most comprehensive long-term care plans are those that are better researched, of known etiology, easily identifiable, or have

greater public awareness (Phillips & Secrest, 2005; Griggs et al., 2009). Better care plans are associated with both increased health-related QoL (HrQoL), and reduced long term healthcare costs. For example, by helping people with diabetes manage their condition effectively, the risk of complications, such as amputation of digits or limbs, can be reduced ("The effect of intensive diabetes therapy on the development and progression of neuropathy"). However, many LTC are not easily identifiable or widely known about, even amongst healthcare professionals, as they may only affect a small proportion of the population. This study was concerned with the Ehlers-Danlos Syndromes (EDS) and examined the role of external support in helping people with EDS manage their condition.

EDS affect people of every ethnicity, are associated with poorer than average health-related quality of life (HrQoL), and higher than average levels of anxiety and depression (Berglund, Pettersson, Pigg, & Kristiansson, 2015). In addition, researchers believe that understanding EDS could lead to a model for research into treatments for other types of chronic pain (Castori, 2016). In particular, investigation of the etiological biochemical processes underpinning symptoms of EDS (particularly those of the extracellular matrix) could offer insight into other LTC. EDS has been described as "a paradigm collagen disorder" (De Paepe & Malfait, 2012, p.1), a view that is supported by a recent review of matrix biology and EDS (Byers & Murray, 2014). Subsequently, research into optimal management of EDS may have implications for provision of support for people with other LTC.

EDS: An Overview

EDS are a group of heritable disorders of connective tissue (HDCT), with several variants (see: appendix 1, table 1). As of March 2017, they are described by an international classification based on inheritance pattern and genetic etiologies (Malfait et al., 2017). The 2017 nosology classified EDS into thirteen sub-types (see: appendix 1, table 3). The introduction of high-throughput genetic sequencing methods has driven research into the etiology of EDS, as well as its phenotypic range of expression, complications, co-morbidities, and disease burden. However, EDS remains incurable and research into comprehensive long-term management of, and coping with, EDS is lacking.

Causes of EDS. EDS are monogenic disorders caused by variants of several different gene loci associated with collagen production (see: appendix 1, table 3). Specific gene variants cause different types of EDS (Malfait et al., 2017). Research has identified causative gene variants for all EDS subtypes bar hypermobile EDS (hEDS) (see: appendix 1, table 3). Recent research considers hEDS to be a heterogeneous subtype, which it is posited may be found to be several distinct subtypes with disparate, and possibly polygenic, etiologies (Zweers, 2003; De Wandele et al., 2013; Lyons et al., 2016).

EDS affect collagen, a major structural protein of the various connective tissues. Collagen deficiencies in the most prevalent forms of EDS are caused by abnormalities of production of collagens I, III, or V (see: appendix 1, table 3). As a result, despite disparate genetic etiologies EDS are expressed in

broadly similar ways, and typically affect skin and joints as well as circulatory and gastrointestinal systems. However, EDS display significant differences between the subtypes due to the specific type of collagen affected and its role in the body.

Signs and symptoms. EDS are variable in expression, however are characterised by joint hypermobility (JHS: joints that move easily beyond the typical range), delayed wound healing, abnormally stretchy skin, easy bruising, and tissue fragility (Callewaert, Malfait, Loeys, & De Paepe, 2008; Grahame & Hakim, 2010; Ericson & Wolman, 2017). These are present in each sub-type, however severity and prevalence differs. Common complications of EDS include chronic pain (Berglund et al., 2015; Scheper, de Vries, Verbunt, & Engelbert, 2015; Castori, 2016), sleep disorders (Guilleminault et al., 2013), fatigue (Voermans et al. 2010b), joint dislocations and subluxations, early osteoarthritis, and gastrointestinal issues (Grahame & Hakim, 2010; Zarate et al., 2010; Zeitoun et al., 2013). Other manifestations include dysautonomia (Tinkle et al., 2009; De Wandele et al., 2014) such as postural orthostatic tachycardia syndrome (Celletti et al., 2017), gynaecologic and obstetric issues (Hugon-Rodin, Lebègue, Becourt, Hamonet, & Gompel, 2016), poor proprioception (Clayton, Jones, & Henriques, 2015); insensitivity to anaesthetics (Wiesmann, Castori, Malfait, & Wulf, 2014), as well as skeletal deformities such as scoliosis, swan neck deformity and tethered spinal cord syndrome (Ericson & Wolman, 2017; Henderson et al., 2017).

Diagnosis and testing. Each subtype has clinical criteria, typically consisting of major and minor criteria, to assist diagnosis of specific subtypes (See: appendix 1, table 1). There is symptomatic overlap both between the subtypes, and between similar conditions. EDS have historically been clinically identified using differential diagnosis and subtype-specific criteria (Beighton et al. 1998). As genetic etiologies were identified, genetic testing for specific gene variants has become possible. Of the subtypes, hEDS as yet has no identified genetic etiology, so continues to be clinically diagnosed (see: appendix 1, table 2).

EDS frequency and phenotypic overlap with similar conditions. The overall frequency of EDS is frequently cited as estimated to be 1 in 5000 (Callewaert et al. 2008). However, EDS-HT is an under diagnosed condition that may correspond with JHS (Castori, 2012), suggesting the overall frequency may be higher than that stated. Castori (2012) stated that the Villefranche Hypermobility EDS (EDS-HT) demonstrates significant phenotypic overlap with JHS, and that EDS-HT and JHS are considered to be the same condition. This concept has been accepted by some researchers (Tinkle et al., 2009), but not by others (De Paepe & Malfait, 2012), although the current nosology combines JHS and EDS-HT into hEDS (Tinkle et al., 2017). Hypermobility, classic and vascular types of EDS are the most common forms, though prevalence of the subtypes established by the 2017 international classification has yet to be established. Furthermore, research indicates a higher prevalence of hEDS, and Hakim and Sahota (2006) suggested a frequency of 0.75-2% JHS/EDS-HT in the general population. Fibromyalgia

shares some common features with EDS, particularly hEDS (Hermanns-Lê & Pierard, 2015; Di Stefano et al., 2016), and studies have suggested a link between the conditions (Ofluoglu, Gunduz, Kul-Panza, & Guven, 2006; Sendur, Gurer, & Bozbas, 2007).

Medical Care and Management of EDS

Broadly, the objectives of medical management of EDS are: to manage the symptoms of EDS and associated complications and co-morbidities; to minimise the possibility of adverse events; and to lessen impairment of QoL (Grahame & Hakim, 2008; Byers et al., 2017). EDS are complex, multisystemic disorders with variable presentation both between and within the sub-types. In order to deliver multidisciplinary care, medical management must be coordinated or overseen by a care team with sufficient knowledge of EDS to understand the wide-ranging implications for HrQoL (Grahame, 2008). Some therapeutic approaches have been assessed in the literature, including significant modalities such as physiotherapy (Pacey, Tofts, Adams, Munns, & Nicholson, 2013), and several researchers have emphasised multidisciplinary management (Grahame & Hakim, 2010; Grahame & Kazkaz, 2014; Chopra et al., 2017). Treatment for EDS is palliative in nature, and centred on symptomatic therapy (Bird, 2007) such as treatment of musculoskeletal symptoms, pain management, and cardiac assessment.

Pain and fatigue are common complications of EDS (Voermans, Knoop, Bleijenberg, & van Engelen, 2010a; Voermans et al., 2010b). The prevailing model used to explain chronic pain is often referred to as the “biomedical

model" (Andrasik, Flor, & Turk, 2005), which views pain as resulting from purely biological factors. Criticisms of this model in relation to chronic pain concerns its inability to account for: chronic pain without identifiable pathology; pathology not associated with pain; disparate responses to treatments; pain refractory to analgesic medications; and the inability of research to establish a strong relationship between pain, functional impairment, and disability (Andrasik et al., 2005; Quinn, Johnston, & Johnston, 2013). In contrast, the "biopsychosocial model" views chronic pain in light of interaction between biological, psychological, and social factors (Jorn, 2015). From the biopsychosocial perspective, individual's experiences of chronic pain and their responses to treatment result from complex interrelationships between biomedical factors such as genetics and history of injury, predispositional, and psychological factors, as well as individual's social and cultural background such as prior learning or beliefs about pain (Andrasik et al., 2005).

Chronic pain management strategies for individuals with EDS typically utilise physical therapy and medications and may also include durable medical equipment such as splints, compressive garments, or braces (Chopra et al., 2017). Physiotherapy and other physical therapies are usually intended to address pain and fatigue, and to assist rehabilitation after injuries. Research shows that issues such as poor proprioception (the sense of relative position of body parts, and movement within joints) and fatigue have a complex relationship with muscle strength and activity limitations (Celletti et al., 2012; Scheper et al., 2016). While muscle strength appears to be a dominant factor in activity limitations, this is confounded by proprioception, suggesting that

physical therapies that also aim to enhance proprioception are pertinent to management of EDS (Rombaut et al., 2011). The current literature suggests the long term efficacy of physical therapies on chronic pain resulting from EDS is lacking (Castori, 2016).

The effects of differing forms of physical therapies on long term management of EDS are also as yet unclear (Castori et al., 2013). Some research has investigated the impact of exercise mechanics on people with HMS with respect to the effect of exercising into hypermobile rather than neutral joint range (Pacey et al., 2013). Exercise into the hypermobile range was reported to benefit child psychosocial health, while exercising to neutral only was perceived to benefit physical health. This suggests that different forms of exercises may be appropriate for different stages of rehabilitation.

Behavioural and lifestyle changes to reduce the risks of injury and pain may be beneficial to optimal management of EDS. For example, injury can be sustained by repetitive microtraumas and abnormal joint loading (Pacey et al., 2013). Interventions, such as occupational therapy, can assess activities and facilitate the adoption of less detrimental methods of accomplishing daily tasks (Sobey, 2014; Ericson & Wolman, 2017). Studies also suggest that the use of orthotics, such as casts, braces, or arch supports, are generally helpful (Agnew, 1997; Bird, 2007), but typically require tailoring. Conservative use of assistive devices, such as wheelchairs, splints, and canes, may be required to facilitate daily function. Assistive devices should be used with caution in order

to avoid decline of supported muscle groups or damage caused by redistribution of load (Ericson & Wolman, 2017).

Other treatment for EDS concerns management of non-musculoskeletal issues, such as gastrointestinal dysfunction, sleep disorders, or dyautonomia. For individuals with vascular EDS (vEDS), cardiac-valvular EDS (cvEDS), and rare subtypes, monitoring for potential complications specific to the subtype is an important aspect of management. For example, it is recommended that people with vEDS are regularly assessed by a cardiologist (Sobey, 2014). Genomic counselling prior to planned pregnancies may also be recommended; vEDS in particular raises the risk of pregnancy complications, while other subtypes appear to be associated with increased risk of spontaneous abortion, multiple spontaneous abortion, and post-natal complications (Hugon-Rodin et al., 2016; Bowen et al., 2017; Brady et al., 2017).

Medication can be used to address various aspects of EDS. Analgesics or medication that addresses neuropathic pain are often the primary factor in pain management (Arthur, Caldwell, Forehand, & Davis, 2016). Research suggests antidepressants, antiepileptics, and opioids may be of use for neuropathic pain while nonsteroidal anti-inflammatory address nociceptive pain (Camerota, Celletti, Castori, Grammatico, & Padua, 2011). Medications alone are likely to be largely unsatisfactory as a long-term pain management protocol and there are adverse effects to long-term use of analgesics (Chopra et al., 2017). Other reported use of medication for complications or co-

morbidities of EDS included cardiovascular and pulmonary medicines, and anti-depressants (Rombaut et al., 2011). EDS are associated with raised levels of psychological distress, and antidepressants may be helpful to address psychological conditions as well as pain management (Bulbena et al., 2017).

Psychological therapies are also relevant to management of EDS. The relationship between EDS, particularly hEDS, and psychological distress is well documented (Lumley, Jordan, Rubenstein, Tsipouras, & Evans, 1994). Research also supports an association with anxiety disorders, depression (Hershenfeld et al., 2016; Bulbena et al., 2017), and neuro-developmental disorders (Ghibellini, Brancati & Castori, 2015). Psychiatric and psychological approaches are recommended to relieve the clinical conditions associated with EDS, and to improve individual's ability to cope with their LTC (Bulbena et al., 2017).

Current treatment for EDS, particularly for hEDS, is typically considered to be insufficient (Chopra et al., 2017). Researchers suggest this may be due in part to an overshadowing focus on musculoskeletal symptoms and subsequent oversight of less obvious, non-musculoskeletal symptoms (Rombaut et al, 2011). Emphasising the need for research into means of improving specific symptoms, Castori et al. (2013) identified that all published treatment recommendations, including their own, were based on the clinical opinions of experts. Therefore the current treatments for EDS are, in the most part, not evidence-based. Several studies have reported that knowledge and

understanding of EDS and hypermobility by medical practitioners is variable and often lacking (Grahame, 2008; Berglund, Mattisson, & Randers, 2010; Sobey, 2014). Research suggests lack of knowledge of rare diseases fuels a 'diagnostic gap', and a lack of evidence-based long-term management options (Bergland, 2014). Research has also reported iatrogenic injuries (injury caused by medical treatment or care) in physical rehabilitation of individuals with hEDS (Bovet, Carlson, & Taylor, 2016).

Coping with EDS. Coping is often broadly differentiated into emotion-focused or problem-focused coping with stressors (Folkman & Moskowitz, 2004). Coping is typically achieved via regulation of emotional response to stressors, or resolution of the problem/stressor (Lazarus & Folkman, 1984). Emotion-focused coping minimises the effects of stressors via cognitions such as emotional regulation or positive reappraisal of the stressor. In relation to EDS or other LTC, emotion-focused coping may take the form of re-evaluation of short- or long-term goals in light of physical limitations. Emotion-focused coping may be most effective in situations that are perceived to be outside the individual's direct control. Problem-focused coping mitigates stress via situational solutions, when the stressor can be influenced by the individual. In relation to coping with stress caused by EDS or other LTC, problem-focused coping could include a proactive approach to healthcare, as an attempt to ensure better healthcare options (Steiner, Bigatti, Hernandez, Lydon-Lam, & Johnston, 2010).

There are few studies that address ability to cope or acceptance of disability (AD) in people with EDS (Bergland, Mattiasson, & Nordström, 2003). Bergland et al. (2003) reported that individuals with EDS displayed similar levels of AD, and sense of coherence to individuals with other LTC. High levels of AD are linked to better HrQoL and general QoL, and so AD can be seen as an important factor in coping with EDS.

QoL and HrQoL. QoL refers to general well-being, and is a concept used in relation to individuals, groups, and societies. While there are different measures used to quantify QoL in literature, they assess life satisfaction in relation to a number of dimensions, which typically include health, education, and standard of living (Hsieh, 2013). HrQoL is a distinct concept that is concerned with the impact health status has on QoL. Measures intended to quantify HrQoL typically assess both mental and physical health, as well as functional impairment (Yin, Njai, Barker, Siegel, & Liao, 2016).

Social Support. While consensus on what defines support remains elusive, theories of social support have posited that support is related to management of stress and coping (Williams, Barclay, & Schmeid, 2004). There are two broad schools of thought concerning the mechanisms by which support is helpful to individuals. Some researchers suggest that it is the emotional component of supportive actions that are salient (Semmer et al., 2008). Others suggest that it is the relevance of the type of support to the characteristics of the challenge(s) faced by the recipient that determines how

supportive the interaction is perceived as being (Cutrona, 1990; Sarason, Sarason, Brock, & Pierce, 1996).

Social support has been investigated in the literature in relation to LTC other than EDS such as: chronic obstructive pulmonary disorder (COPD: Arabyat & Raisch, 2015); heart failure (Heo, Lennie, Moser, & Kenedy, 2014); breast cancer (Gonzales, Hurtado de Mendoza, Santoyo-Olsson, & Nápoles, 2016); diabetes and depression (Coffman, 2008); as well as adherence to treatment in hypertension (Magrin et al., 2015). While many studies on support in relation to LTC do not isolate the types of support as distinct constructs, in general the literature supports the notion that emotional support is linked to improved emotional well-being, improved physical symptoms and reduced depressive symptoms (Heo et al., 2014; Gonzales et al., 2016), and thus to improved HrQoL. However, Heo et al. (2014) observed that individuals with depressive symptoms may perceive support differently to individuals without depressive symptoms, which they suggest may imply that alleviating depressive symptoms as well as improving emotional support should improve physical symptoms and HrQoL. Emotional support from friends and family has also been shown to have a relationship with adjustment to disability, as did self-esteem (Li & Moore, 1998). A study on the effects of tangible support, depression, and diabetes reported a relationship between lack of support and patterns of nonadherance to treatment and poor self management (Coffman, 2008).

Magrin et al. (2015) examined functional social support, which they operationalised as comprised of emotional, instrumental and informative social support. Their meta-analysis found that functional support is related to increased adherence to treatment for hypertension; however they noted that there were insufficient studies that examined sources of support as distinct constructs to isolate their effects with respect to hypertension. There appears to be similar issues with research into other LTC. For example, while Arabyat and Raisch (2015) noted a link between support and depressive symptoms in line with other research findings, they aggregated emotional and social support in their study. While different forms of support, or measures of forms of support, are held to be highly correlated in the wider literature on social support (Sarason et al., 1996; Semner et al., 2008), the tendency of researchers to aggregate types of support currently complicates attempts to understand the mechanism(s) associated with differing types of support. Despite these methodological issues, the literature suggests social support has a complex, multidimensional relationship with health, via psychosocial functioning and health-related behaviours such as adherence to treatment.

There is limited research on support and EDS, although one study reported that coping and support predicted QoL (Fox, 2014). The researcher stated that support was a strong predictor of QoL, and suggested that medical interventions for pain management should incorporate support and coping techniques. This corresponds with other research that substantiates a relationship between social support, coping and health (Uchino, 2006; Segrin & Passalacqua, 2010).

Self-management support (SMS) is a recently defined concept in health management that encompasses comprehensive, long-term strategies intended to improve outcomes for people with LTC (Kawi, 2012). These strategies involve patient-focused factors such as involving individuals in their own care, provision of pertinent information in an accessible manner, and tailored care. They also involve factors related to care-provision, such as being an informed provider, possessing appropriate skills, and cultivating favourable attitudes to facilitating care and self-management (SM). A final factor to SMS consists of organisational attributes such as incorporating social support with care plans, having the structure in place to facilitate multidisciplinary care, and an organised system of care for LTC (Kawi, 2012). Researchers have stated that SMS has been identified as a priority for providers with respect to LTC such as chronic pain (Lukewich, Mann, VanDenKerkhof, & Tranmer, 2015), JHS (Terry et al., 2015), and diabetes (Song et al., 2012). Effective SM is linked with better QoL with respect to several LTC, including COPD (Moriyama, Takeshita, Haruta, Hattori, & Ezenwaka, 2015) and diabetes (Kueh, Morris, & Ismail, 2017).

There are peer support groups for people with EDS. Several are funded and hosted by charities such as EDS UK, whereas others are organised by their members. Some support groups have in-person meetings and thus are composed of people who live proximate to one another. Others are facilitated by social media and can have international membership. The advantages of online support include access to more individuals with EDS, ease of communication, and on some platforms anonymity, which some individuals

may find preferable when discussing sensitive or personal issues (Coulson, Buchanan, & Aubeeluck, 2007).

Research into effects of support on coping with, or optimal management of, EDS is extremely sparse and merits further investigation. This study aimed to answer the research question: 'how does external support help people with EDS manage their condition?'

Method

This was a qualitative study which gained full ethical approval from the University of Chester's Ethics committee. Five female participants took part. All participants were volunteers who were not offered inducement/s for participation in the study. Participants' age ranged from 20 to 53, with a mean age of 33.6 (*SD* = 14.2). All participants were adults who had received a diagnosis of any subtype of EDS, more than twelve months prior to recruitment. Time since diagnosis ranged between 2 and 7 years, with a mean of 3.6 years (*SD* =1.9). This study was concerned with experiences and perceptions of support available to people with EDS. It was assumed that participants living with an EDS diagnosis for more than one year prior to recruitment have experienced the full range of support available to them, some of which may only have become available after diagnosis.

Using a convenience sampling frame, participants were recruited from online EDS support groups hosted on social media sites, by word-of-mouth, or by responding to posters placed in a North-West University and in a North-

West public library. All participants were members of online support groups and resided in the UK.

People interested in participating in the study were advised to contact the researcher for further information. Potential participants were sent the participant information sheet (PIS: see appendix x) and invited to take part in an interview. Interviews took place via video call or in a private room in a North-West public library. Before the interview participants were instructed to read the PIS again in full, and asked to sign the consent form (see: appendix x). Participants were advised that audio recording would begin when the interview started. Interviews were between 26 and 70 minutes in duration, mean interview time was 42 minutes (*SD* = 17.1). All interviews were audio recorded using a handheld digital recorder. Participants were asked to read the debrief package before leaving or ending the call. Data collected was subsequently transcribed for analysis from the recordings using an orthographic transcription notation system (Braun & Clarke, 2013). Participants were assigned code numbers that were also used in transcription to preserve anonymity. Interview text that identified locations, participants or other persons was redacted from the transcripts.

Thematic analysis was then carried out using the anonymised transcripts. The researcher gained initial familiarisation with the texts during transcription and redaction. The texts were re-read several times and familiarisation was finalised by marking every piece of text that appeared to be significant, prior to coding. Initial coding was carried out on text identified as

significant. Initial codes were generated using participant's terminology, in order to retain both surface and contextual meanings, as well as to begin to organise codes by relation to one another. As interviews were coded, reoccurring topics, and consequently related codes, became discernible. When initial coding of all interviews had been completed, several topics salient to the participants had been identified. At this stage a master-list of all codes generated in initial coding was compiled and conceptually similar codes were reduced, as appropriate. Initial coding was then repeated on the transcripts to ensure all significant discourse had been identified and tagged. Coding was deemed to be completed when no new information was emerging from the text and data saturation was achieved.

Codes were then grouped; the groupings were found to be most coherent and relatable when grouped by support source, so the main sources of support were identified as themes. Subthemes were then identified by grouping tags within those themes by salience rather than prevalence, although subthemes that had been mentioned by most or all participants were noted. Once the comprehensive list of candidate themes and subthemes was compiled, the transcriptions were re-examined to verify that all text of significance had been identified and that the themes identified could be meaningfully applied to all interviews. The themes were then reviewed and consolidated for clarity, and four themes with 11 subthemes were identified. The themes identified prompted further subject research, which informed thematic definition and naming.

Reflexivity

My experiences of support for students with disabilities informed my expectations of institutional support, while prior familiarity of personal experiences with medical care for EDS will have influenced my understanding of medical support. Additionally, I was familiar with peer-support groups before inception of the project. These experiences influenced initial research and first order coding, however unanticipated themes emerged in second order coding that prompted thorough recoding to saturation, and largely informed later research. Some analysis was influenced by my prior knowledge such as: an understanding of the salience of stigmatisation to people with visible or invisible stigma; and understanding of the nature of institutional support for students with disabilities.

Findings

Four themes were identified; most were comprised of several subthemes. Theme one, community, included the subthemes: spouse; family; friends and acquaintances; and peers. Theme two was institutional support. Theme three, medical support, included the subthemes: diagnosis; the role of the GP; types of care; and optimal care. Finally, theme four, the role of self-support, included the subthemes: resilience; lack of resilience; and the role of support in the development of resilience.

Theme 1: Community

The theme community was defined as the (lack of) support from external sources. While some of the types of support available may also be available through medical sources, community sources were conceptualised as being distinct from medical sources due to the disparate nature of those relationships. Community sources are not presumed to have had a medical background and support-provision is typically less formalised. This theme included the subthemes: spouse; family; friends and acquaintances; and peers.

Subtheme 1.1: spouse. The subtheme included any romantic partnership. Several participants suggested their spouses were a primary source of physical/tangible, and/or emotional support. When reflecting on support received from her partner, one participant stated that he was physically supportive to the point of relocating dislocated joints, and was emotionally supportive by being tolerant of the participant's negative moods.

> "my boyfriend as well, he's <u>really</u>, wel- because I've been with him like five years. So he's been with me since I've had this diagnosis, so he knows the drill, he knows how to put certain joints into place if I can't quite ah, he puts up with the (.) strops ((laughs))" (1001, p5, 30-34)

Her spouse's willingness to provide such support on an as-needed basis suggests tolerance of the variability of EDS.

Community sources such as spouses may be motivated to emotionally support an individual with EDS, but lack aptitude, competence or confidence to do so.

> “the sort of emotional side of it, I a-, it’s kind of between me and my partner, but I think he then gets very frustrated because he doesn’t like know how to help me in that way” (1004, p11, 20-24)

The participant acknowledged the difficulties inherent in trying to provide emotional support that her spouse may not feel able to provide, and the impact this may have. Participants further suggested that developing LTC is also stressful to the spouse and can alter the nature of the relationship post-onset of EDS. One participant suggested that the heritability of EDS, lifestyle changes and increased financial responsibilities can be serious stressors of spousal relationships.

> “when we got married [several decades] ago, I had a bad back ((laughs)) an a- and a wonky ankle, I- I didn’t have EDS with the likelihood I was going to pass it on to any children that we would have. And erm, he’s a good man, and he has taken it you know but, it has impacted us it’s impacted the life we had ... impacts lifestyle that impacts, the sharing of the load as it were financially for our family, it impacts, who he is to me and our relationship” (1005, p10, 19- p11, 5)

Several participants referred to physical/tangible support from spouses such as sharing household tasks. One participant reflected that the support her spouse gave her enabled her to live independently.

> “my partners really good, like, he, like we live together, it’s just us two and, the dog ((laughs)) and um, like he, kinda knows really, there’s a lot that I can’t do around the house and things, so he, again, they’re quite physically, supportive and that helps me, live with him” (1004, p4, 25-p5, 3)

Another participant cited her spouse as a primary motivator for maintaining resilient attitudes and behaviours.

> “You know, there’s some days and I’ve not wanted to get up, and every day I have to think ‘right, come on, let’s get up’ an- and I do it for [husband] as well as myself because If I just stayed in bed depressed that’s not fair on him either.” (1002, p16, 6-10)

Subtheme 1.2: family. This subtheme focuses on the role of family members in supporting people with EDS. One participant stated her family provided her with near-daily physical support.

> “quite often they’ll nip in most days and check that I’m OK and, erm, whether I need any sort of physical help, erm, to do anything and, like, if not they’ll ring me, every day and, same with my grandparents when [parent]’s away, erm, so knowing that I’ve got that, physical help if I need it” (1004, p3, 15-22)

Daily contact with family members, or other close social relationships, may also reduce feelings of isolation and/or vulnerability in people with physical limitations. However, although family can provide tangible and also emotional support, the participant expressed concern about the negative impact of providing such emotional support on the family.

> “I think my worry for me is, I don’t wanna put it all on her because she’s still struggling, with the fact that I’m like this and need all that help, nobody wants their child to be in that situation an- It causes her a lot of anxiety me being like this and, I don’t want to put it all on her, like, as in, ‘you be the sole support for me’” (1004, p11, 13-20)

She implied her concerns caused her to constrain the support accepted from her mother, in order to moderate negative emotional impact. Another participant felt there was a lack of support for, and understanding of the impact of EDS on immediate family members. The participant felt responsible for helping them cope and adjust to her condition.

> “our daughter of course ... all her life all she’s known is Mummy to be in pain or to be having surgeries, and, there’s been massive impact on the pair of them, outside of my control outside of my own, ability to know what to do to help them ... for me, that’s as important getting that right, getting them, I don’t know a safe place, to sound off or be upset or, ask ‘what now?’, at the moment that’s all come from me too” (1005, p11, 5-20)

In this context, support for immediate family members is support for the person with EDS. Support for family could relieve people with EDS of perceived responsibility for the emotional impact of EDS, as well as by reducing stress to those relationships.

One participant reflected on her parents' attempts to educate themselves about EDS. She inferred that understanding of EDS facilitates belief and trust in the person with EDS.

> "my parents have made a lot of effort to understand about EDS an, and (.) the kind of, biology behind it all and everything (.) So they really do believe me, if I say something rather than just saying 'oh it's another thing'" (1004, p8, 32- p9, 3)

This was salient to the participant as she stated that being believed by family members is essential to having enough confidence to seek support. Family and friends who display understanding of EDS were cited as a primary source of support.

> "having, friends and your family that understand completely everything, and they know that sometimes you're gonna have bad days, when you're not going to move about, but there's going to be great days when you can go and do anything, at all ((laughs))" (1001, p10, 28- p11, 2)

It was the understanding of the variability of EDS that was appreciated by the participant. She implied that this enabled her to make the most of times when her symptoms were less severe.

However, the nature and quality of support from extended family was perceived to be variable. One participant felt this was due in part to their own non-resilient attitudes such as denial or avoidance of the implications of the heritable nature of EDS.

> "it's a very patchy, sort of erm, response of support in the wider family, I think partly because some of them are fearful that they've got it and they don't want it, and they've seen what I've been through, erm and some of them it's a- head in the sand you know 'it might go away if I don't think about it'" (1005, p14, 28- p15, 7)

Subtheme 1.3: friends and acquaintances. This subtheme focused on support from friends and acquaintances. One participant reflected that there were limitations to support from friends due to the nature of these relationships.

> "with my friends I've not gone on and on about the condition but they've been aware of how I struggle with it. I've, you know I have to, take a step back and think 'I can't go on and on about it because it's not fair on them', and it will take over all of my life, so if they ask me how I am, I- I just tell them, you know, 'it's not nice and I struggle' and, but then I go onto something else" (1002, p14, 25- p15, 2)

The participant seemed to want to find a balance between conflicting desires surrounding her friendships. She wished to discuss EDS with sufficient frequency and detail that friends were aware of its effects, making them more

likely to be understanding of her limitations. Nonetheless, she wished to protect her friends by reducing the negative emotional impact of conversations about EDS.

Most participants cited willingness to tolerate and adapt to limitations as a key feature of valued friendships. This participant reported that there was one individual she felt particularly close to, who would collaborate with the participant to ensure group activities were inclusive.

> “I’ve got one friend in particular who, I sort of confide in quite a lot. And we always make sure that whatever we’re going out to do is suitable, and make sure that everyone else is aware that you know, things might happen ((laughs)) things are just going to happen.” (1001, p5, 18-23)

By initiating conversations with others in the friend group, the participant’s confidante may have reduced misunderstanding and negative cognitions about the participant’s limitations. One participant contrasted a lack of understanding and an attitude of disbelief with that of people who do believe her when she described symptoms.

> “people always used to say to me growing up ‘there’s always something wrong with you’, or ‘you’re always not very well’ and, you were kinda like ‘yeah, but I don’t know why’, and, start to question yourself as whether it’s real or, I- ((exhales)) ‘why doesn’t everybody else feel like this’ but nobody understands what it’s like because nobody else feels

like this and then, t- to have people who do, believe you when you're saying that all those times" (1004, p9, 9-18)

She suggested that being doubted by others can cause individuals to doubt themselves and question the veracity of their own experiences. Such self-doubt can hinder access to support, as individuals must first accept their need for support in order to access support.

Participants implied that support was important to successfully adjusting to and coping with diagnosis or onset. This participant cited a close friendship that she credited with making her feel less isolated.

"I think having her there has made a big difference to me kind of, getting through this, emotionally, and, knowing that I still have people around me" (1004, p16, 31- p17, 2)

The repeated references to loss of relationships and expressions of gratitude regarding sustained friendships implied participants had underlying insecurities about close relationships. This suggests fear or anxiety regarding potential lack of social support. For example, one participant viewed close relationships as being essential to coping with severe adversity.

"I am very lucky and I don't know how people on their own (.) Cope with something what turns their life upside down" (1002, p15, 30-32)

Subtheme 1.4: peers. Peer support encompasses support from others with EDS via peer support groups. Some of those interactions took place

online, others at in-person group meets. Other instances of peer support involved family members with EDS, as well as friends outside of a peer group setting. All participants mentioned peer support groups, most reported positively on their experiences.

> “there’s also the support with sort of the online groups, sort of with the facebook groups and umm the instagram community, the instagram spoony community is absolutely amazing, when I- when I first found out every- (.) Yeah ((laughs))” (1001, p1, 20-25)

Several participants spoke of the peer group’s role in dissemination of knowledge & information about EDS. One participant reported that she valued the wealth of accumulated knowledge the groups represent. She detailed her experiences with an auxiliary treatment, which she had found to be beneficial, and that she advised her peers to try.

> “the different Facebook groups? Yeah absolutely amazing yeah, we’d, we just, all know so much, like I know the miracle cure for me is Epsom salt baths, that’s like, the nearest thing to a miracle, and I’ve sold so many I should be on commission” (1002, p11, 21-28)

Another participant valued the local knowledge that peer groups disseminate. She recounted use of the online peer groups to seek peers experiences of specific medical professionals or services.

> “The only reason I go on to, the message boards is if I’ve got a particular question ... I was seeing a particular rheumatologist and I just

> put out there ‘anybody seen this rheumatologist?’ ‘What is he or she like?’ ‘What do I- what do I need to take with me?’ And that was helpful.” (1005, p5, 3-10)

A participant suggested that peers offer a different kind of understanding and support, based on insight derived from lived experiences.

> “I think a lot of the people that are around you can support you but they don’t necessarily understand you, or- yeah, um, that support from somebody who, understands what you’re going through, cos I think, n- as hard as, say your parents or my partner and that try, they don’t quite get what it’s like to live with it every day, so, it’s a differen- although the people like that I speak to online might not be as close friends (.) them having an understanding offers quite- a more unique sense of support” (1004, p2, 17-26)

Feeling comprehensively understood by a person who has similar lived experience differs from understanding gained from researching an illness. It is likely that interactions with peers are less fraught with uncertainty, compared with interactions with people who do not have EDS. Conversely, another participant reported negative perceptions of peer groups; she stated that she was angered by dissemination of misinformation, as well as propagation of non-resilient attitudes.

> “I spend very little time on the, chat rooms and that sort of thing because, actually, it makes me quite angry, that there’s a lot of ill

> informed, wittering, and ‘oh woe is me!’ as opposed to ‘I’ve got this, I don’t like it, how can I make my life better?’ which is absolutely my way of looking at life.” (1005, p2, 1-10)

She perceived the groups to be focused on validation and comfort, whilst being avoidant of challenging or critical discussions.

Theme 2: Institutional Support

The theme institutional support was identified. This included support from groups or organisations where the support received was not on a person-to-person basis, such as NHS provision of specialist services.

One participant reflected on the potential impact of organisations’ lack of understanding about EDS.

> “when it comes to work places, or erm, educational institutes, especially if y-, if you’re having to have a lot of time off, erm, they don’t necessarily understand, why you are ill all the time, which can be such a big barrier and it can make you feel very depressed and a bit like, ‘well maybe-, maybe I’m not worth doing that job or maybe I can’t do that job (then) maybe I should just quit, maybe I should just give up’.” (1003, p2, 17-26)

Lack of understanding in institutional support can inculcate negative self image, which may foster mental ill-health and non-resilient attitudes. This can lead to people with EDS living more restricted lives than necessary.

Several participants reflected on their experiences of tangible/physical support from educational institutions. Such support facilitates study by reducing some of the physical burden on students with EDS.

> “at university I had quite a lot a lot of support when I first went it was sort of um (.) chairs and help with note-taking, and, umm (.) I was given printers and stuff to help me so that I didn’t have to walk down to the library and print stuff off at uni, I could print, at home.” (1001, p1, 33 – p2, 4)

Provisions of this nature can help mitigate extra challenges that students with LTC face. Other non-physical accommodations were also positively reflected upon, such as extensions to deadlines.

> “it’s a hard enough time for anybody let alone, when you’ve got so much going against you in a way like, jus- just knowing say, you’ve got extensions on your work takes off that bit of pressure” (1004, p18, 14-18)

Students with LTC may experience more occasions when they are prevented from working at their full capacity by health issues. By facilitating greater flexibility about assessment deadlines, universities can mitigate additional stress experienced by students with disabilities at such times.

Several participants spoke of issues pertinent to institutional support as it related to medical care. One participant felt that the NHS could facilitate peer

groups, and thus social support, for people with broader health issues such as chronic pain.

> “I think at the doctors it would be good if they did have support groups for patients with chronic long term, health conditions ... you know I think whether it’s EDS. you’ve got, whatever, you know is really affecting your day to day quality of life, it would just be a really good, support network and a lot of people don’t have family and friends. You know, so that could be a real lifeline” (1002, p2, 17-29)

Another participant stated that medical institutions could play a role in dissemination of accurate, evidence-based information.

> “things like, you know, the NHS websites, I know they are, better now it does mention EDS. Erm, but you know, again, if all them are kept up to date with you know, off the HMSA and EDS, erm, you know, with what’s happening” (1002, p4, 15-21)

Most participants spoke of issues relating to resource limitations within the NHS. One participant stated that people with EDS would benefit from comprehensive multidisciplinary care. She wondered if there was a relationship between lack of NHS resources and scarcity of such care options.

> “I dunno if it’s because NHS is under-funded, under-resourced, er overworked, but it needs to be a much more of a holistic approach, t- to everything” (1003, p21, 11-14)

Another participant spoke about her experience of a specialist, multidisciplinary clinic, and stated that such care would be more beneficial if proximate.

> "Well I've just been to [specialist orthopaedic hospital] in [month] for a three week rehab on chronic pain, which, because of all the cuts and whatnot, that's in [large southern city]. You know, wouldn't it be amazing to have one in the North?" (1002, p5, 17-22)

With the lack of proximal support, participants were willing to use other physical therapies that are more locally available. One participant reflected on her experiences of a council-funded scheme.

> "we have an exercise referral scheme, for people that've got disabilities ... that was really helpful because I went and I-, the lady actually understood, what my condition was, which, she was able to, tailor a gym package, and exercise like regime which didn't put too much impact on my joints, but allows me to build the muscles, and build the core stability, which was really good" (1003, p11, 5-23)

Such specialist schemes can foster habitual resilient behaviours, such as regular physical activity.

Another issue related to institutional support was lack of consistency of care for EDS in the NHS. One participant contrasted the lack of standard care for EDS with care received by family members for other heath issues.

> “I think there needs to be an EDS diagnosis pathway ... if somebody is diagnosed with erm, if I think in my own family, multiple system atrophy or, non Hodgekins lymphoma, or in my case heart failure, you’re on a pathway. You’re on a pathway th- the NHS has the, guidelines the next steps the pathway laid out, the increase of diagnosis of EDS the sort of very few experts that there are nationally that recognise it and can deal with it to who people are referred, means that it’s all very much more piecemeal, there isn’t a pathway yet” (1005, p37, 6-21)

The participant felt that a care pathway should facilitate the development of resilience in people with EDS. She acknowledged individual differences in psychological adjustment to diagnosis or onset, and stated that a care pathway could offer chances to help individuals who struggle to develop resilience unsupported.

> “for people who, don’t necessarily have the ability to think ‘right ok, what support do I need?’ I think, there needs to be, a guide path- a pathway, certainly a suite of, issues and opportunities that can be offered, to help people get through and become the best of themselves with EDS that they can be.” (1005, p42, 7-12)

Finally, most participants referred to EDS charities, which host many of the online peer-to-peer groups, and facilitate local support groups. One participant identified the charities as a source of information about EDS. She suggested that someone newly diagnosed should look for personal accounts for information on individual differences in EDS presentation.

> “Get on the EDS UK website, get all the information packs, erm, get on the internet and read the stories that are ‘my life with EDS’ cause they’re all very very different” (1005, p33, 28-31)

One participant stated that the charities and the support groups they host have been a source of support for family and friends of people with EDS.

> “Well the support groups I go to are the HMSA, they have a theme every month, and one month a year is ‘family and friends’ so you invite your family and friends to talk to other people with EDS, and it gives them, obviously an insight” (1002, p5, 1-5)

Participants deemed information was critical to understanding, and understanding of EDS meant support-providers, such as family, were better able to support people with EDS. Subsequently information support for friends and family of individuals with EDS can facilitate community support.

Theme 3: Medical Support

The theme was conceptualised to encompass (lack of) support from healthcare-providers. This included medical care from disparate disciplines such as physiotherapy and psychology. This theme included the subthemes: diagnosis; the role of the GP; types of care; and optimal care.

Subtheme 3.1: diagnosis. All participants spoke of diagnosis as a critical factor in medical support. Most mentioned delayed and misdiagnoses. One participant reflected on her delayed diagnosis of EDS after childhood

onset of symptoms. She reported a misdiagnosis of growing pains (GrPn) resulting from her reports of wrist pain. This would have been atypical for contemporary understanding of GrPn.

> “When I was first trying to get diagnosed, when I was first, getting the first sort of symptoms, it was mainly in my wrists, because I was sort of younger I s- went to the childhood (.) hospitals, and (.) they passed it off as growing pains for about three four years” (1001, p3, 33 – p4, 5)

All participants who spoke of GrPn suggested they felt the diagnosis of GrPn to be inappropriate, or misleading in relation to children who presented with early signs of EDS.

> “I just hope that the next generation, are still not going to the GPs and they say ‘it’s growing pains’ and you know, this and that” (1002, p16, 19-20)

One participant reported inappropriate, invasive treatment due to misdiagnosis of gallstones when she had costochondritis, a co-morbidity of EDS.

> “I was having severe chest pains, and I was told ‘oh no it’s just because you’ve got gallstones, we’ll take your gallbladder out, it’ll be fine’ Ok ((inaudible)). They took my gallbladder away, I’ve <u>still</u> got chest pains, it’s all to do with the, I can’t pronounce it, it’s- it’s erm, it’s an EDs condition, um EDs associated condition and it’s, the muscles between the ribs spasm .... So I had to have this very invasive procedure, I had

> to have nearly a month off work, because of, of the procedure itself, and it didn't actually remedy the problem" (1003, p21, 16- p 22, 3)

Misdiagnoses could also be the result of disbelief of reported symptoms.

> "the children's hospital where they just kept putting it off as growing pains, that was the bit where the wait was, the disbelief, that something was wrong" (1001, p14, 4-7)

When another participant reflected on her experiences of disbelief, she stated there was a paradox in the relationship between belief and diagnosis. Once individuals are diagnosed with EDS, they are more likely to be believed, however, in order to be diagnosed, they must first be believed.

> "when you don't know what's wrong, it's kind of like, well (.) they kind of, doubt you in a way as to whether this is happening all the time as much as you're saying it is, erm, yeah, whereas I think when you do have, a diagnosis, of something (.) people believe you more, ((laughs)) but, to get that diagnosis you need people to believe you, an it's this, kind of, catch twenty two of, 'how do you get help without knowing what's wrong?' yeah, erm, and I find since I've had a diagnosis, the support's been, much, better, from everybody, because it's like 'well now we know what's wrong', 'there is something wrong' ((laughs)) 'we do need to help you'" (1004, p10, 5-27)

She noted that diagnosis also facilitates support from other sources. Several participants stated that lack of knowledge of EDS in the healthcare-providers was problematic, as diagnosis relies on understanding of EDS.

> “Well, it took me, fifty years to get diagnosed, so, the aware- the lack of awareness widely in the medical profession, of this condition, is a massive barrier” (1005, p27, 5-9)

Another participant related that she was not given information about EDS post diagnosis. She stated that lack of information hindered the provision of support by family members.

> “when I found out that I did have it, that was it, I was told I had it, and left in the dark. So none of my family knew about it, so none of my family knew how to support me” (1001, p8, 15-20)

One participant related how her diagnosis led to early diagnosis of EDS for her child. When reflecting on her symptomatic history, she stated that early diagnosis will enable her daughter to make choices that should have better long-term health-related outcomes.

> “I’ve had [many] orthopaedic surgeries, I’m looking at quite a few more cause I’ve got rampant arthritis caused by the traumatic injuries to all my joints over the years, she doesn’t have to have that, she doesn’t have to have that, if she sets (.) she sets herself up now” (1005, p8, 20-28)

Early diagnosis would mean her child has more choices than she did, however; her child must make them. She had expressed concern that this may limit her child's life choices and experiences, and reflected on how early diagnosis might have affected her decisions with respect to her career and reproductive choices.

> "I've often thought if I'd had my diagnosis in my twenties, would I have lived the life I have done? Would I, have chosen to have our daughter? ... would I have, stopped doing the job I passionately love to do something that was less physically demanding?" (1005, p12, 21-28)

Subtheme 3.2: the role of the GP. General Practitioners (GP) were the primary source of medical support reported by participants. When reflecting on her pain management regimen, this participant referred to how her GP facilitated her minimalist approach to analgesic medication.

> "I do get sort of the pain relief side of things from my GP... it's quite helpful, but because I'm on the tramadol, and I can't take codeine, and I can't take um, the anti-inflammatorys, I can only take the pain relief, when it's got to the point where I can't cope any more" (1001, p6, 15-25)

By supporting ad hoc usage, rather than prescribing a broader medication regime, the participant's GP enabled her to be responsive to day-to-day variability of pain symptoms and in control of her use of medications.

GPs are the point of contact with the healthcare system and can grant access to specialist treatment. One participant stated that GPs facilitate life-long care, which supports the notion that the relationship with a GP is a critical one within the context of medical support.

> “I think, for me, a- a big help i- is your GP, in that, they’re kind of like, the port of call for all of your medical and psychological help like, they can, send you to where you need to go, at any point throughout your life” (1004, p14, 11-16)

This participant stated that people with EDS had a role to play in fostering beneficial relationships with GPs.

> “for me information is power. Knowledge is power ... You have to be responsible for yourself, if you want your GP to help you in the future they need to understand what it is you’ve got, and odds are you know more than they do, and it certainly was the case certainly was the case for me.” (1005, p34, 6-21)

Another participant expressed empathetic feelings about how GPs may feel when treating patients with LTC. She alluded to different care approaches, implying that a palliative rather than curative approach was beneficial to people with EDS.

> “It must be frustrating for them cos as a doctor, you want to make people better, and because they can’t it must be very frustrating” (1002, p8, 32–33)

The palliative approach can ease disease burden by addressing symptoms that may exacerbate other issues, such as sleep disorders worsening chronic pain.

Another participant reported feeling discouraged when she had an unsupportive GP, and more proactive when she had a better relationship with a GP.

> “now, I feel like I can go, and I will get some support, whereas, when I was kinda-trynda-find-a, a better GP, I was like, I was going, an it was, felt like a waste of time, and like nobody was listening, and that, there was no support. an- I think it makes a difference of feeling like, you’re either on your own dealing with this, or, you’ve got the backup of somebody who’s willing to help you, live with it” (1004, p5, 28- p6, 5)

Subtheme 3.3: types of care. Participants spoke of different disciplines relevant to people with EDS. Of those, physiotherapy and psychology emerged as particularly salient. There were polarised views on physiotherapy, for example, iatrogenic injury from inappropriate treatment was reported.

> “the first physiotherapist I saw, was absolutely wonderful knew loads about EDS taught me loads about how to treat EDS ... he transferred me onto a different physiotherapist that was working at the same place, and he was absolutely awful. And within the first five minutes, my knees

were dislocated, my shoulders were dislocated, didn't have a clue (.) how to go about it" (1001, p3, 1-12)

One participant was critical of incomprehensible or unrealistic advice she received regarding physical rehabilitation.

"I'd had physios then tell me 'well you just need t- to walk with soft knees', right, it's not that easy ((laughs)) it really isn't when you learned to walk like this, you've literally got to retrain your brain how to wor- how to walk, and that's not something you can do with a physio session of twenty minutes once a month." (1003, p14, 29- p15, 6)

In contrast, one participant reported more beneficial treatment was delivered by physiotherapists who displayed an understanding of her physical limitations.

"my new physio is not too bad actually, he- he understands a little bit more, and he knows that it's an ongoing thing, so he's spread my physio out a lot more than previous physios have ... it means you've got longer to try and get used to the exercises, he understands that I can't, do (.) ten exercises a day every day, because he knows that he ((inaudible)) i- it's just not not physically possible, he s-, what he, what he tries to do is he tries to give me two or three exercises that I can do in my everyday" (1003, p6, 30- p7, 12)

In spite of concerns about the potential for misguided physiotherapy to result in iatrogenic injury, and/or impede their progress with physical

rehabilitation, participants stated that physiotherapy was an important type of care for people with EDS.

Psychological treatments were spoken of by most participants. One participant suggested that her knowledge of EDS and coping mechanisms was not adequate by itself. She stated that support from her psychologist enabled her to apply this academic knowledge to herself.

> “I’ve done a degree in [social science] and I’m doing my Masters in [healthcare-related science]. So, you kinda learn about all these things but, I never really applied it to myself, it’s like, you’re learning it for oth-, to do with other people, and um-, it kinda, took someone to kinda say ‘you need to, think about yourself as well in the same way’” (1004, p11, 30- p12, 9)

This suggests that objective external support can assist the development of resilient attitudes by challenging assumptions about coping with EDS. Another participant stated that psychological support can benefit people with EDS during the post diagnosis adjustment process.

> “You also need to have, some form, of counselling too, (because?) when I first got diagnosed, you suddenly don’t-, it hits you like a brick wall that you can’t necessarily be able to do everything, you wanted to do. Erm, because you’re kind’ve, grieving for the life you won’t get now, and that’s-, hits some people a lot harder than others” (1003, p16, 17-24)

One participant suggested that psychological support can free people with EDS from perceived responsibility for the emotional impact of provision of support on support-providers.

> “it’s, <u>really</u> beneficial to hav- have somebody, to go to like you say that’s objective that, isn’t tied to you, emotionally, and that you can feel like you can go and, and speak to about anything and not upset them or not worry them or, ((laughs)) and they’re, trained to help you with that as well.” (1004, p19, 30- p20, 8)

In this context, psychological support may be seen as having reduced the emotional burden on community sources of support.

Subtheme 3.4: optimal care. Participants spoke of care that was either tailored to people with EDS or delivered by specialists in a field pertinent to EDS, multidisciplinary care, and comprehensive treatment. While one participant was reflecting on support from friends, she observed that she had better QoL when she was receiving appropriate medical care.

> “when I first met them I didn’t know about my diagnosis, I didn’t know about my condition, I didn’t know how to manage it, and (now) they’ve seen the progress in me and the change in me as well, especially when they’ve got medication right, because then my moods change, I’m much happier when I’m there and, that kind of thing, and they’ll know, if they see me on my crutches that I’m having a really bad day and, they, try and help me out however they can” (1003, p23, 12-21)

This implied that as she had become better informed about her condition and management of EDS, and begun use of assistive devices when needed, that others in her group had become more aware of her condition which informed their provision of tangible support. Thus, incidentally, optimal care had also facilitated community support.

One participant implied that EDS-specific care, or greater awareness of the implications of EDS within specialisations, could have had a beneficial effect on her management of EDS.

> “Now when I’m trying to go and explain stuff there’s-, there’s things that I don’t think are related, to EDS and whatnot, and what I need support for, but they actually are, related, but if I’d had that support in place from the start that would’ve just been a natural ‘oh, this happened, this happened, this happened’, rather than, think back years and years and years, to god knows what” (1001, p10, 6-15)

Had this participant received EDS-specific care she could have been assessed for related conditions post-diagnosis. As she did not receive this sort of care, the participant was addressing these issues several years later when symptoms had become pronounced enough to warrant referrals to specialists.

Understanding of similar LTC can help ensure non EDS-specialist care is appropriate, responsive to the needs of people with EDS and effective. One participant related positive experiences with a team of therapists who had an understanding of similar collagen deficiencies.

> “the hand therapist I saw actually had hypermobility herself, it wasn’t- she wasn’t EDS, but she was just, had hypermobile s-, a few hypermobile joints, but, it meant she knew a lot more about it and she’d been able to educate her team ... Um, it really helped, it really helped because of, because she managed to educate them, they kind of, saw it from our side of view as well, and didn’t just treat it as if I’d hurt myself, it was an ongoing thing so tha- that the package of the exercises were made, and tailored, to an ongoing condition” (1003, p5, 5-18)

Symptomatic treatment represents the palliative rather than curative approach to management of EDS. This was illustrated by another participant’s account of her experiences of multidisciplinary care, which reduced the disease burden of co-morbidities of EDS by improving functioning.

> “most of my problems are with my knees, being very loose, but I know that could be because of my pelvis is loose but my upper body is not too bad, erm, bladder issues, I did have problems, but because I’ve done the correct physio and sit-ups, they’re ninety odd percent better now. Erm, I’m lucky I can eat what I want, I don’t have any stomach issues. My POTs issues I’ve gone on slow sodium so they’re nin- abou- eighty five ninety percent better” (1002, p17, 5-18)

Several participants mentioned multidisciplinary clinics as an example of optimal care. One participant observed changes to her friend’s management of EDS after attending a multidisciplinary clinic. She suggested that the

multidisciplinary nature of the clinic led to her friend gaining a better understanding of EDS, and developing resilient behaviours.

> "cos they've got that multi-agency linking, they've got that working there. It's-, they've got the counsellors there, they've got the specialists there, they've got physios there, they've got dieticians there, and they all work together, when she came out of there, yes she came out with another like four diagnoses, but, she also come out understanding the condition a lot more, and knowing where her limits are." (1003, p24, 20-28)

Another participant related how modelling what was done at the multidisciplinary clinic had resulted in her incorporating other daily activities into her management of EDS.

> "I learnt a lot being at [specialist orthopaedic hospital] every morning at nine o clock, they used to put erm chillout CDs on and do stretches for half an hour. That was a really lovely start to the morning so I've bought two CDs for at home, erm and I do <u>that</u> erm, and that's been a massive help" (1002, p18, 1-7)

One participant felt that having received optimal (i.e. comprehensive multidisciplinary) care, she had enough knowledge to manage her EDS in her current state.

> "I see a rheumatologist at [specialist orthopaedic hospital], erm, and then I see cardiologist, fairly close by, um, gastroenterologists as well,

> in [city]. Trying to think. I was seeing like physios and OTs and things, but, we kinda got to the point where it's, I've got as much knowledge as, as possible really" (1004, p7, 4-11)

As a congenital condition, progression of which typically involves chronic complications, optimal care requires ongoing treatment.

> "I think what can really help is when you've got a doctor that's willing to listen, ((inaudible)) and willing to accept that EDS is a chronic illness, it needs help constantly, it's not some -at that they can (.) just forget about and then it's going to be fine." (1001, p7, 14-17)

Being an informed patient does not exclude people with EDS from other health issues throughout their lives. This participant estimated that her condition changes around every 18 months, and onset of severe symptoms of EDS was precipitated by pregnancy.

> "things sort of carried along very nicely until I fell pregnant and then everything changed then, and then it's about every eighteen months, about every eighteen months now, where something, new happens ((laughs)) shockingly, yeah" (1005, p13, 24- p14, 2)

Theme 4: The Role of Self-Support

Self-support encompassed attitudes and behaviours deemed likely to affect management of EDS, and HrQoL. Optimal self-support was contrasted with attitudes and behaviours that resulted in poor management and of EDS.

Several subthemes emerged related to the notion of self-support: resilience; lack of resilience; and the role of support in the development of resilience.

Subtheme 4.1: resilience. The subtheme resilience conceptualised behaviours and attitudes deemed characteristic of effective management of EDS. Resilience was perceived to be related to better long-term health-related outcomes. One participant stated that resilient attitudes enabled access to specialist medical support that helped her manage EDS effectively.

> "I'm very stubborn and a fighter and I will, I wanted to go to [specialist orthopaedic hospital] and I got there but, when some people are dealing with so many issues, they can't fight, everything can they? ... an- and then they their health can deteriorate can't it" (1002, p10, 10-17)

The participant described the process of learning to manage EDS as inclusive of several stages or factors; psychological adjustment to diagnosis, developing understanding of EDS, disseminating relevant information to friends and family, and acceptance.

> "Well, because it is sort of a self management program we have to do there's no miracle cure, umm, it's coming to terms with your diagnosis, um, learning to get your head round things, mentally and physically, umm and communicating it to loved ones, and family and friends, which you know, we do feel sort of angry and g- go through different emotions umm when you're in chronic pain, umm, and at the end of it- it's

> acceptance and until we get to that point, it's just getting through the day ((laughs)) as best you can every day." (1002, p1, 16-26)

Resilience was also perceived to be learnt in childhood as one participant suggested her resilient attitudes were due in part to her upbringing.

> "I think I- I'm quite different from a lot of people in that regard I suppose because, the way I am and the way I was brought up was just to crack on with life rather than to give in" (1005, p1, 30-32)

Resilient attitudes were perceived to have a complex relationship with pain management. This participant spoke of the psychological adjustment she experienced subsequent to having experienced a several week residential rehabilitation course at a specialist orthopaedic hospital.

> "it's a big reality when you get home and the pain hasn't gone, ((laughs)) you know and it's, you know even though you've had the pain for a long time, to get your head round, god, every single day I'm on this earth I'm going to have to deal with this and it's going to be tough" (1002, p6, 31- p7, 3)

In the context of development of resilience being a process rather than an innate and static quality, her statement is indicative of a key stage in this process. Adjusting to, and accepting EDS and resultant limitations, is necessary in order to develop a long-term view, and make informed choices that may improve HrQol. For example, another participant had reported adverse reactions to medication. She stated that since these adverse

reactions, her most effective option for pain management was a positive attitude.

> “it’s almost a psychological approach to it, it’s, ‘today is going to be OK, unless my body tells me otherwise’.” (1005, p24, 12-14)

One participant related her mental fortitude to her choice to engage in therapeutic physical activity despite high pain levels. Furthermore, she linked her mental fortitude to management of anxiety.

> “if your anxiety is high, it can take years, like my- I’m sat here now and my pain, not this particular minute, but my knees are looser than they’ve ever been, but yet, I can go to bed and sleep straight away when years ago I had panic attacks, didn’t sleep, I couldn’t eat, erm, and just over time I’ve thought I’ve got this, erm and I’m not going to let it, you know ruin my life ... just having belief and, strength that you can do it every day and setting yourself goals and challenges and going out of your comfort zone, I still go swimming, I go to the gym, erm, you know and some days I’m crying and in agony and I can hardly walk, but I still go. So I am stubborn” (1002, p18, 15-32)

Her statement was indicative of her long-term focus with respect to management of her condition. Another participant expressed how development of resilience related to acceptance of limitations imposed by EDS. This facilitated acceptance of support, which is essential to accessing support.

> "I did my first few years without support because I was (.) I'd only just been diagnosed I didn't, think I needed it, but as time went on, I was getting worse I really did need, much more support and accepting I needed that support was, a big step, especially after being so independent" (1004, p18, 3-10)

Self-reliance was reflected in statements about EDS management, and maintenance of boundaries to do with external support. One participant demonstrated resilience by statements indicating she was able to weigh her desire for independence against her need for external support. Her acceptance of the effects of EDS tempered her self-reliance, which allowed her to reject external support when unneeded, and seek it when necessary.

> "sometimes I just have to say 'look I'm not great but I'm OK', like, umm, 'I'm gonna be alright and I'll ask you if I need some help'" (1004, p4, 21-23)

Feeling smothered by inordinate support may add to feelings of frustration and prompt negative thoughts/loss of self-worth. Consequently support is helpful when needed and wanted, and may be deleterious to well-being when superfluous.

Most participants referred to actively seeking out information about EDS, which was deemed to confer several benefits. Information was validating and reduced self-doubt, thereby boosting self-esteem and confidence.

> “I’ve read a book ... that covers every single aspect of daily living with the condition, <u>everything</u>, so I would recommend that to people ... if someone had’ve given me that book when I was diagnosed an- I’d, an- I, you just think ‘god, it’s as though I wrote this’. You know you’re just reading and thinking, ‘they get it’ (1002, p11, 6 – p12, 7)

One participant stated that information about EDS allowed her to make sense of symptoms of co-morbid conditions.

> “all the different parts of my life that have always just been ‘oh that’s [participant’s name]’, actually are connected, Erm like my hideous inability to deal with hu- humidity for instance it’s PoTS, now I know what it is” (1005, p3, 3-10)

Understanding co-morbid conditions can be essential to management of those conditions, and thus contributes to better HrQoL. When reflecting on the comparatively high level of medical support she had received, the participant suggested that her communication skills had benefitted her in her relationships with healthcare-providers.

> “I’ve been really lucky, but also (.) I communicate hard, I (.) I make sure I’m, I- I know what I’m talking about, and I ask questions” (1005, p19, 32- p20, 2)

A key behaviour characteristic of resilience is a proactive approach to one’s own care. She further stated that for her, resilient attitudes such as self-

reliance, as well as good communication skills, had a relationship with resilient behaviours such as a proactive approach to healthcare.

> "Why would you give your power away to a faceless doctor or a GP who sees five thousand other people a year or anybody else, why would you do that? Why would you not take responsibility for yourself, to A be informed, B engage in a conversation with your primary medical provider usually your GP, and then be, proactive about making the best of the situation, why would you go, 'oh it's all in the GPs hands' or 'I have to deal with the GP to get this sorted' and-, I don't know why you wouldn't take responsibility for your own body." (1005, p26, 12-20)

One participant expressed the belief that her diagnosis occurred after she began to assert herself, and proactively sought out specialist medical care capable of providing explanations for her ongoing issues.

> "they passed it off as growing pains for about three four years, until I s- I actually stuck my foot down I was like 'no I want to see a specialist (.) and I want to get this-, I want to find out what's happening', because I was seeing the same doctor every time for those three years, and they were not having it what-so-ever." (1001, p4, 5-12)

Assuming a proactive approach to one's own care as early as possible was deemed critical to establishing comprehensive medical care.

> "get to the GP and, try and get referred to the different people that you need to get referred to, erm ((laughs)) start sorting out everything

straight away, cos that's the thing that I wish I'd been able to do is get everything sorted cos now it's so hard being so far into it and trying to get anyone to listen." (1001, p9, 27-33)

As detailed, resilient attitudes foster resilient behaviours. However, one participant also suggested that performance of resilient behaviours can foster resilient attitudes.

"at the end of the day I've got to write down like sort of three good things about the day (.) and whether that's practical things or whether that's like the other areas like 'well I just managed to stay awake all day' that's ((laughs)) that's a big thing for me an-, At first I was like 'oh, you know I know these things' but, I didn't really, think about those things as being good things and like to have them all written down and, you know to see what I ((inaudible)) am still doing or, the good things about me that day, is-, is really helping me I think, like, trying <u>not</u> to focus on the things I haven't done." (1004, p13, 18- p14, 1)

Subtheme 4.2: lack of resilience. Lack of resilience collated attitudes and behaviours that were not characteristic of resilience, and which were perceived to have a negative relationship with HrQoL and/or successful management of EDS. Passivity was identified from behaviours such as a passive approach to healthcare. One participant stated that a passive approach to her own care had resulted in her not receiving appropriate care in a timely manner.

> “don’t do what I did, and assume the doctor’s got it covered. Push and push and push, because I naively just thought ‘well the doctor’s sent the referral off I’ll just wait for them to do their bit’. No, and er two years later I’m still waiting for my referral.” (1003, p15, 19-26)

One participant inferred a passive approach to health care can result in individual’s health care being deprioritised or delayed by medical support providers.

> “a lot of people that you, meet on the message boards, are all ‘ugh I don’t know what’s happening’, or ‘is it normal to wait this long?’ Get on it! You know, be proactive, make the phone calls make a nuisance of yourself, erm, because the people who sit around waiting, are, not going to go to the front of the queue that’s just how it is” (1005, p5, 19-26)

She also noted a relationship between passivity and negativity, which she felt online groups were overly tolerant of.

> “I don’t necessarily see that with a lot of, <u>other</u> fellow EDS people, who I see on the notice boards taking pictures of a dislocation and going ‘what do you think this is ladies?’ it’s a fucking dislocation, go and get it sorted. Sorry, but do you know what I mean it’s-, it- it’s all, bollocks when you start turning in on yourself, and feeling sorry for yourself.” (1005, p20, 4-15)

One participant reflected on poor management of EDS, which she linked with lack of knowledge as well as behaviours and attitudes characteristic of lack of resilience. She referred to a cycle of deconditioning, which can exacerbate pain and worsen physical limitations.

> “a lot of it is about, keeping up with your physio and n- as, you know, as fit as possible but, when you’ve got a body what can dislocate and you’re in a lot of pain, people can be frightened to, you know to do things like that, because they think ‘am I making it worse’, and, and as well with th- all, with all the anxiety side of it a lot can’t even you know, they don’t feel confident to go to support groups and, talk to other people, um, and then you end up in a spiral, the more pain you get, you stop doing things, and i- i- it just go, it can go down in a spiral” (1002, p9, 24-31)

Another participant also reflected on deconditioning, and observed that the cycle can be interrupted via physical therapy.

> “Get yourself, some form of physical therapy that works for you whether it’s physio, osteo, pilates, swimming, just do it. Because no matter how bad you are you <u>can</u> get better, <u>than</u> <u>you</u> <u>were</u>, you will never be perfect, but there is this cycle you can get into, and it’s happened to me, where pain leads to deconditioning, deconditioning leads to more pain, pain leads to increased deconditioning. And I have been there, and it’s cyclical” (1005, p35, 4-15)

Denial of EDS is discernible in statements suggestive of avoidant behaviours. This participant reflected on avoidant behaviours exhibited by some of her siblings.

> "One of my siblings is a GP, clearly has it [himself/herself], will not discuss it. Will not discuss it ... I have a[n other sibling] who is amazing, who, I believe has got it, I believe all [their several] kids have got it, but [they've] never asked me the question and it's not for me to say 'go and get yourself, looked at'. Erm, there are, very clear signs that [he/she] has got it." (1005, p14, 16-28)

Avoidant behaviours have thus precluded this participant's siblings from seeking diagnoses, treatment, or any other form of support for issues the participant believed were strongly suggestive of EDS. Other avoidant behaviours include resistance to appropriate use of management options such as analgesic medication, assistive devices or institutional concessions. This participant made several statements concerning her reluctance to use such options. Her resistance in considering herself to be 'disabled' meant she had delayed application for institutional support to which she was entitled, and which proved to be beneficial.

> "for years people have said to me 'you should get a blue badge [participant's name] you know you should just get a blue badge' I'm like 'no no no!', 'that's not me I'm not disabled cos I hate that word' blah blah blah, and then [several] months ago I did get my blue badge, and it's been wonderful, it has truly helped." (1005, p9, 13-17)

Subtheme 4.3: the role of support in the development of resilience. Participants suggested that support can encourage resilient behaviours. One participant related that she would suggest people newly diagnosed utilise institutional or peer support groups to access information.

> "I'd probably suggest getting onto one of the support sites straight away, just so that you've got that massive background knowledge of different things and different opinions on where you can go, who to go to, who to speak to, what it could be, what it might not be, and sort of having that back-up and having people who know exactly what you're going through and have been in the same place, it, encourages you a little bit, makes you feel a lot more supported" (1001, p9, 15-23)

A participant related how a residential course at a specialist orthopaedic hospital helped her develop resilience. The participant described provision of comprehensive healthcare, dissemination of multimodal information about EDS management, and development of understanding about pain management.

> "It was amazing, it was absolutely life changing for the better, yeah, um, to be on a ward with twenty thirty other people, um three quarters of the people had the condition, erm, and you've got your own psychologist, occupational therapist, physio, you cover anatomy, how the body's built, how it works, about medication, about diet, foiling flare ups, covers absolutely everything, what pain is, how it goes up your spinal cord and tells your brain that you're in pain, and basically it's a programme to

> learn you that pain's in the driving seat and you're in the back seat and it's about you being in the driving seat, and pain being in the back seat, you're acknowledging what's happening, but it's how your brain processes it." (1002, p6, 8-25)

One participant suggested that support in general should foster a proactive and positive attitude to management of EDS rather than dwelling on the condition, and thereby failing to adjust psychologically subsequent to diagnosis.

> "support for me is, a common sense (.) 'how do we make life better?' 'how do we move forwards with this in the knowledge this is what it is?' rather than a wallowing in" (1005, p4, 28-29)

The participant also suggested that psychological support would be beneficial to help people with EDS develop resilient attitudes and approaches to pain management. She suggested medical support to enable people with EDS to undertake physical conditioning while reducing risk of injury. She stated there was a relationship between failure to develop resilience in relation to pain management, and poor health-related outcomes.

> "some degree of psychological support, particularly around pain management, because, I- for me pain management is now the key, and that's what I took to the rheumatologist, erm yes I've got problems with my hand and I need an injection but actually, the key was, <u>pain</u> management, and I- I know what my limitations on painkillers are I know

> what I can take and can't take, so what the next steps all around that are, the (.) occupational therapy, slash, psychological support that goes with, making sure you don't injure yourself but you do keep pushing yourself, and don't run away from the pain because I know a lot of people run away from pain and it actually works, much more, negatively, than it does if they were to confront it" (1005, p36, 14-27)

Discussion

The present study examined the role of external support in helping people with EDS manage their condition. The themes conceptualised issues concerning (lack of) support from disparate sources: community; institutions; medical; and the role of self-support. Community incorporated personal or face-to-face support (e.g. spouse, family, friends, and peers). Medical support included specific professionals and types of care available to people with EDS. The role of self-support included and focused on resilience and how support helps development of resilience.

This study reported differing forms of support. These comprised: emotional, which included inclusion or a sense of belonging; physical or tangible, which included assistance with activities, as well as infrastructure or specialist resources; and informational, which included provision of information to participants and to others. The effects of these forms of support have not been investigated in relation to EDS. However research into the efficacy of forms of support has been carried out in relation to other LTC and health issues, including several pertinent to management of EDS.

Types of Support

Emotional support. Unlike research into other LTC, the results of the present study did not explicitly link emotional support with depressive symptoms (Arabyat & Raisch, 2015). However, the results strongly implied that emotional support, from a variety of sources, helped participants to cope with EDS. Emotional support was linked with coping, in terms of 'getting through' the experiences of declining physical condition, initial lack of medical explanation for physical decline and other symptoms such as pain, subsequent diagnosis of EDS, and resulting lifestyle change(s). Individuals who were credited as being participants' main source(s) of emotional support were also described as the primary motivators for participants to resist patterns of thought and behaviour characteristic of depression, as well as of lack of resilience. Further, those individuals cited as primary support sources were also credited with being participants' motivation to engage in resilient behaviours such as regular physical activity. Such behaviours are likely to have been beneficial in improving symptoms of depression and anxiety (Dinas, Koutedakis, & Flouris, 2011), as well as potentially easing some symptoms of EDS, and protecting physical functioning (Chopra et al., 2017; Engelbert et al., 2017). Participants' also credited of emotional support-providers with making them feel less alone or vulnerable. Subsequently the results of this study suggest mechanisms by which emotional support has a relationship with depression.

Tangible/physical support. Participants reported that they found tangible/physical support useful in a number of ways; physical support was reported to facilitate independent living by assisting with necessary daily tasks, facilitating medical care, and enabling social interaction. Research into tangible support and other LTC largely corresponds with these findings. A study of the effects of tangible/physical support and depression on diabetes self-efficacy similarly reported that spouses and family were participants primary source of support and that their support facilitated healthcare, enabled independent living, and was associated with adherence to treatment (Coffman, 2008). Studies have reported tangible support enabled people with LTC to develop achievable goals and adopt coping strategies, as well as noting that too much support can hinder the development of resilience (King, Willoughby, Specht, & Brown, 2006). The researchers suggested this may result from inordinate support preventing the individual from adopting coping strategies that would enable them to complete tasks themselves. This echoed the findings of the current study. The subtheme: resilience reported that unasked-for help was deemed unhelpful, and participants preferred support that allowed them the independence to do what they are capable of.

Informational support. The results suggest that informational support is particularly salient to individuals with EDS. This is congruent with research suggesting information is essential to ‘empowerment’ of people with rare diseases, which has been reported to have a relationship with HrQoL (Aymé, Kole, & Groft, 2008). Informational support facilitates individuals becoming knowledgeable about their condition, which is essential to make informed

choices about management of LTC (Aymé et al., 2008). Individuals with EDS must make informed choices about care/treatment options, which their EDS may have had implications for, including medication and viability of surgical options (Wiesmann et al., 2014). Participants reported iatrogenic injuries and unnecessary treatment, which they implied was less likely to have occurred subsequent to participants having become more knowledgeable about EDS. Further, lack of knowledge of EDS in healthcare-providers was cited as a causative factor of iatrogenic injuries and unnecessary treatment, which is supported by similar findings in research (Grahame, 2008; Berglund et al., 2010; Sobey, 2014).

The need for information to facilitate support was reported in relation to all four themes. This may be partially attributable to the complexity of EDS, as well as the relatively low awareness of EDS by the general population, including medical professionals (Grahame, 2008; Berglund et al., 2010; Sobey, 2014). Information on EDS was reported to be essential to institutional support which is typically tangible/physical in nature. The results of this study illustrated how institutional support can facilitate academic success by mitigating some of the extra burdens and negative cognitions (for example, concerning attendance) that students with disabilities experience. It is also possible that support for students with disabilities facilitates adaptation to student life and influences integration with healthy peers by encouraging access to local health resources (Herts, Wallis, & Maslow, 2014).

Sources of Support

Does the source of support alter the impact of types of support? Pertinent to research concerning support is Thoits' (2011) observation that the impact of support types may be mediated by the source of support. Thoits suggests that support from "experientially similar others" (Thoits, 2011, p145), or peers, may be perceived differently by individuals. The current study reported on support from different sources in ways that implied congruency with Thoits' observations; participants spoke of concerns about impact of provision of support on family and friends, and the limitations of support from community sources. Participants also spoke of peer support differently to support from medical or community sources, and suggested that peers experiential understanding qualified their opinions on pertinent issues such as proximate healthcare options, auxiliary treatments, or coping strategies. This is consistent with Thoits' (2011) argument that support provided by experientially similar others was perceived differently to support from community sources such as spouse or friends, and also congruent with the observation that constructively critical support may be highly beneficial from peers, but be perceived as 'unsupportive' and elicit negative emotional reactions when received from spouses (Romano et al., 1995).

Peer groups. The current study highlighted peer groups as a source of uniquely qualified support. People with HDCT frequently report feeling isolated, misunderstood, and abnormal (Baeza-Velasco, Gély-Nargeot, Bulbena, Vilarrasa, & Bravo, 2011). The subtheme: peers, reported that peer

groups provided individuals with EDS with a sense of feeling oneself to be understood, which suggested that a shared social identity, based on similar experiences of EDS, normalises the experiences of individuals with EDS. This normalisation may counter feelings of abnormality and attendant negative cognitions. This is congruent with research that stated that contact with patient groups may be useful in relation to feelings of isolation and marginalisation (Baeza-Velasco et al., 2011).

Peer friendships have been shown to correlate with well-being in other LTC (Silverman, Molton, Smith, Jensen, & Cohen, 2017). Individuals with more friends who shared their diagnosis were reported to display attenuated associations between the severity of impairment, QoL, and social role satisfaction, suggesting that their peer friendships buffered the impact of their functional impairment on well-being. Further, the study reported similar benefits for individuals with more friendships with people with any disability, rather than a shared diagnosis (Silverman et al., 2017). This suggests that some of the benefits of peer support groups may not be same-diagnosis specific, but may extend to peer groups consisting of individuals with similar LTC. Consequently, peer support groups for less specific health issues may be beneficial to individuals with EDS, which was also a finding of this study, and formed a part of the theme: institutional support. Broadening the scope of peer groups should result in increased memberships of local groups, which could improve access to proximate peer support in less populated regions. A drawback of this approach may be decreased access to experience-based information about specific LTC, which is reported by the current study and

others as a key benefit of support groups (Subramaniam, Stewart, & Smith, 1999; Coulson et al., 2007; Delisle et al., 2016). However, support groups may be able to address that deficit by facilitating online access to information about LTC.

This study also reported some potentially detrimental effects of peer groups, such as dissemination of misinformation and encouraging maladaptive behaviours and attitudes that did not promote resilience. These concerns were supported by research into the benefits and disadvantages of peer groups for LTC such as chronic pain (Subramaniam et al., 1999), chronic muscular pain (Steihaug, Ahlsen, & Malterud, 2002), and rare diseases (Delisle et al., 2016). Subramaniam et al. (1999) reported that peer groups could develop a disempowering focus similar to results reported by the current study that peer groups may focus on validation to the exclusion of discussion of optimal management strategies, or challenging maladaptive behaviours such as passivity, which comprised part of the subtheme: lack of resilience. The above mentioned studies also reported benefits from peer groups such as: availability of information about LTCs; exposure to new coping strategies and resources; modelling of beneficial health-related behaviours; meeting others with similar experiences; emotional support, including validation and recognition; and being able to speak freely about LTC.

Family and community support. Participants of the current study cited spouses and immediate family as primary sources of support, which was also reported in relation to other LTC such as chronic kidney disease (Silva et al.,

2016), and diabetes (Coffman, 2008). The results of this study suggested that ill-health and change in circumstances due to EDS were stressors to those relationships, and can impact family-members well-being, which has also been reported in relation to other LTC including chronic pain and fibromyalgia (Chun, Turner, & Romano, 1993; Mercurio-Riley, Lee, Chronister, & Swigar, 2013; Steiner et al., 2010). Research suggests that spousal relationships may be stressed due to role strains experienced by the spouses in response to factors such as increased household responsibilities, decreased leisure time, and financial strain (Steiner et al., 2010). This is in accord with the theme: community, which reported relationship stressors including the impact of increased financial responsibility and care-giving. Participants reported improved spousal support and reduction of strain to relationships following receipt of external support, such as information support, which improved participants' ability to communicate with, and support, their spouses. Steiner et al. (2010) studied the effects of social support on the healthy spouse, including support from the spouse with the LTC. The effects reported correlated with this study's finding that spousal relationships improved when communication by participants improved, or when participants took responsibility for provision of emotional support for the healthy spouse. Consequently, understanding of EDS must encompass understanding of the effects of EDS on spouses and immediate family members in order to address the stressor effects of a parent or spouse's LTC.

Healthcare-providers.

Diagnosis. The subtheme: diagnosis was deemed highly salient, as diagnosis was reported to be necessary for individuals with EDS to access medical support, and facilitated support from other sources. Most participants spoke of delayed- or mis-diagnoses that they attributed to lack of knowledge or of EDS, which accords with research that suggested a 'diagnosis gap' was driven by lack of knowledge of rare diseases (Bergland, 2014). This was congruent with research into other LTC that has reported delayed diagnoses attributed to lack of understanding of the LTC, or disbelief of reported symptoms (Terry et al., 2015; Armentor, 2017). Studies have reported that disbelief by healthcare-providers can result in distrust and aversion towards medical practitioners in people with LTC; that these attitudes persist; and can impact the quality and nature of care received (Berglund et al., 2010). Consequently, the findings of this subtheme had broad implications for provision of medical support, and suggested that disbelief reported in other themes, including community and institutional support, could also potentially foster lack of resilience, as well as having a negative impact on adherence to treatment or engagement with healthcare.

Finally, accurate and timely diagnoses are pertinent to optimal care for EDS, as intervention in HDCT, as early as possible, may greatly improve HrQoL (Bluestein, 2017; Chopra et al., 2017). For example, in relation to management of EDS, early intervention/s may be critical to reducing disease burden with respect to chronic pain (Scheper et al., 2015; Castori, 2016). This

was reflected in the subtheme: appropriate care, which reported complications from failure to address issues relating to EDS in a timely manner, as well as reports from the subtheme: diagnosis on the advantages to others with EDS who were diagnosed earlier.

Lack of knowledge of EDS in healthcare-providers. Grahame (2008) stated that medical practitioners appeared to lack knowledge of HDCT, and were frequently unaware of recent literature concerning complications of EDS. This supported the current study's findings concerning healthcare-providers who appeared to display a lack of knowledge of EDS, and of complications of EDS. This implies that such lack of knowledge is a major barrier to individuals with EDS accessing medical support, including appropriate and effective treatment. This interpretation accords with similar findings in relation to JHS/hEDS (Terry et al., 2015). For example, when speaking of the efficacy of physiotherapy for HDCT, Grahame (2008) stated that treatment may be ineffective or damaging by being either not aggressive enough, or too aggressive. This is congruent with participants' accounts of iatrogenic injuries caused by overly aggressive physiotherapy or programmes that were too fast-paced. In contrast, therapies that did account for participants' limitations, as well as complications of EDS, were reported to have been highly beneficial.

Psychological care. In line with expectations of the biopsychosocial model, psychosocial and psychological issues were noted by participants as highly pertinent to management of EDS, and it was reported that QoL was affected by psychosocial issues as well as physical issues. This is congruent

with a literature review relating to rare genetic conditions (including EDS) and QoL, which noted that studies that measured psychosocial factors, such as psychological well-being, reported strong relationships with QoL (Cohen & Biesecker, 2010). Further, the subtheme: resilience reported participants' perceptions that medical support for chronic pain related to EDS is largely inadequate, which is supported by research concerning EDS (Rombault et al., 2011; Terry et al., 2015; Chopra et al., 2017)

Self-management and self-management support. Recent research into management of chronic illnesses has highlighted and clarified the concept **of** SMS (Kawi, 2012). The findings of this concept analysis are supported by the theme: the role of self-support, which reported that support can facilitate resilient behaviours and attitudes. The concept of SMS suggests means for healthcare-providers to incorporate approaches that foster resilience into their clinical practice. For example, lifestyle changes and physical therapies can have beneficial effects on coping with, and management of, EDS (Chopra et al., 2017; Engelbert et al., 2017). However, they require strict treatment adherence to be effective. The biopsychosocial model suggests that self-efficacy beliefs would have an impact on individuals' ability to cope with LTC, such as chronic head pain (Andrasik et al., 2005), and may affect individuals' abilities to adhere to treatment plans despite pain. The concept analysis of SMS suggests that by fostering resilience, particularly improved self-efficacy, SMS can encourage increased adherence to treatment, which should result in better HrQoL. This notion is supported by the findings of the current study, which reported increased treatment adherence subsequent to utilisation of

support to address psychological symptoms, and facilitate adoption of a positive and determined attitude.

An example of exemplary care. All participants reflected on a chronic pain clinic at a specialist orthopaedic hospital, which was reported to have combined multidisciplinary medical care with informational and peer support, and a focus on SMS. Participants observed improved management of EDS, in themselves and others, as a result of attending the clinic, which was attributed to several factors. These included learning resilient management behaviours such as activity pacing, becoming more informed about EDS and chronic pain, and that being more knowledgeable enabled them to communicate better with significant others, which resulted in improved familial support. As such, this clinic represents a model of exemplary care for people with serious limitations from EDS.

Currently, such specialist multidisciplinary services are not distributed throughout the UK, and individuals with EDS may benefit from access to similar but proximate services (Grahame & Kazkaz, 2014; Chopra et al., 2017). Early, multidisciplinary medical interventions intended to support SM should result in better HrQoL throughout the lifespan of people with EDS (Kawi, 2012). Consequently, similar services could be beneficial for individuals with less severe symptoms as well as those with severe EDS-related dysfunction. Further, effective SM should result in reduced use of medical resources over time, and thus reduce costs associated with treatment of EDS (Kawi, 2012; Panagioti et al., 2014; Lukewich et al., 2015).

The role of self-support and resilience. Resilience and lack of resilience were key components of the theme: the role of self-support. Statements made by participants suggested that resilience is not a static quality; that development of resilience post-onset or post diagnosis is a process. Attitudes and behaviours characteristic of resilience were implied to be learnable and improvable. Conversely, resilience was also viewed as a seemingly innate predisposition; in other words, a quality of personality that can be present in individuals whether or not they have a LTC, or in the individual with EDS prior to onset or diagnosis. However, literature suggesting resilience is not associated with personality traits is compelling (Roth & Hertzberg, 2017). This is also supported by research suggesting that resilient behaviours and attitudes can be learned and much like other skill sets, such as coping strategies, can be improved (Davydov, Stewart, Ritchie, & Chaudieu, 2010; Schetter & Dolbier, 2011). However, the seemingly contradictory results of this study could be representative of the notion that capacity for resilience may be partially determined by factors such as: genetics, health, and environmental effects (Cameron, Ungar, & Liebenberg, 2007; Conner & Zhang, 2007), and that improvement of resilience occurs within individuals' ranges of capacity (Conner & Zhang, 2007; Davydov et al., 2010). This interpretation is in line with the expectations of the biopsychosocial model.

In line with the theme: the role of self-support, there is support in literature for the notion that individuals' attitudes and behaviours can affect the course and impact of medical care. For example, attitudes and behaviours

deemed characteristic of resilience included inquisitiveness and a proactive approach to one's own care. It appears from research carried out by Fisher (1993: as cited in Werner & Malterud, 2003) that individuals can greatly affect treatment decisions by asking questions. This supports this study's finding that individuals taking a proactive approach to their own care could affect the type and nature of care made available. With the observation that support could facilitate development of resilience, which is reported to be related to coping with LTC such as chronic pain (Newton-John, Mason, & Hunter, 2014), the theme: the role of self-support may therefore represent several mechanisms that may help elucidate the relationship between external support and management of EDS.

Limitations of the current study, and future research. All participants were recruited via support groups. Membership of such groups can confer benefits relevant to management of EDS, such as greater exposure to information about EDS, management of EDS, and availability of care and other support. Therefore, this participant group may be self-selected for high resilience. Further, as participants were reporting on support retrospectively, the results may be skewed towards the impact of perceived social support rather than received support. By diversifying recruitment sources, future studies into the role of support in optimal management of EDS may be able to recruit participants more representative of a range of abilities to manage EDS. There is a paucity of both longitudinal and randomised controlled trials of treatments, support, and management strategies for EDS. There are also many areas of research required to develop evidence-based treatments for

EDS, for example, clinical trials to assess the effects of specific treatments on chronic pain in EDS, such as physical training, CBT, or multidisciplinary approaches. Mixed method or quantitative studies further to the present study, and similar exploratory research, can elucidate the relationships between support, development of resilience, and improved HrQoL for people with EDS. Stratification studies may also allow for more detailed examination of factors that affect management of EDS to determine common elements to optimal management.

Conclusion

EDS are complicated, multisystemic disorders that can impact almost every aspect of an individual's life and severely affect functioning. While research has elucidated the etiology and natural history of most subtypes of EDS, research into optimal long-term management is still sparse and recommended medical treatments are largely based upon the clinical opinions of healthcare-providers. The precise impact of different types of support on EDS remains unclear, and warrants further study, but appears to correspond with the effects of support on other LTC. The results of this study suggest that external support can facilitate the development of resilient behaviours and attitudes in individuals with EDS. SMS is a relatively recent concept in medical care, but could be a promising approach for healthcare-providers to incorporate support of development of resilience into clinical practice. Interventions that incorporate appropriate, multidisciplinary, medical care with the goals of improving individuals' access to support, such as information and peer

support, could enable better management of EDS. Optimal management of EDS is likely to result in better HrQoL and reduce healthcare-related expenditure associated with treatment of EDS.

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Appendices

Appendix 1. Tables

Table 1. The Most Prevalent Subtypes of the EDS, Their Former Nomenclature, and Diagnostic Criteria

2017 International Classification	Villefranche Classification	Berlin Classification	Major Criteria	Minor Criteria	Minimal Criteria suggestive for subtype*
Classical EDS (cEDS)	Classic EDS	types I and II	1. Skin hyperextensibility and atrophic scarring 2. Generalised joint hypermobility (GJH)	1. Easy bruising 2. Soft, doughy skin 3. Skin fragility (or traumatic splitting) 4. Molluscoid pseudotumours 5. Subcutaneous spheroids 6. Hernia (or history thereof) 7. Epicanthal folds 8. Complications of joint hypermobility (e.g. sprains, luxation/subluxation, pain, flexible flatfoot) 9. Family history of a first degree relative who meets clinical criteria	Major criterion (1): skin hyperextensibility and atrophic scarring, plus, either major criterion (2): GJH, and/or: at least three minor criteria
Vascular EDS (vEDS)	Vascular EDS	type IV	1. Family history of vEDS with documented causative variant in COL3A1 2. Arterial rupture at a young age 3. Spontaneous sigmoid colon	1. Bruising unrelated to identified trauma and/or in unusual sites such as cheeks and back 2. Thin, translucent skin with increased venous visibility 3. Characteristic facial	Family history of the disorder, arterial rupture or dissection in individuals less than 40 years of age;

			perforation in the absence of known diverticular disease or other bowel pathology 4. Uterine rupture during the third trimester in the absence of previous C-section and/or severe peripartum perineum tears 5. Carotid-cavernous sinus fistula (CCSF) formation in the absence of trauma	appearance 4. Spontaneous pneumothorax 5. Acrogeria 6. Talipes equinovarus 7. Congenital hip dislocation 8. Hypermobility of small joints 9. Tendon and muscle rupture 10. Keratoconus 11. Gingival recession and gingival fragility 12. Early-onset varicose veins (under age 30 and nulliparous if female)	unexplained sigmoid colon rupture: or spontaneous pneumothorax in the presence of other features consistent with vEDS. Testing for vEDS should also be considered in the presence of a combination of the other “minor” criteria.
Hypermobile EDS (hEDS)	Hypermobility EDS	type III	See: table 2	See: table 2	Clinical diagnosis of hEDS requires the presence of criteria 1, 2, and 3 (table 2)

* Confirmatory molecular testing is obligatory to reach a final diagnosis for all subtypes except hEDS

Major Criteria, Minor Criteria, and Minimal Criteria quoted from Malfait et al. (2017, p 11-17).

Table 2. Diagnostic Criteria for hEDS

hEDS Criteria	Criterion 1	Criterion 2	Criterion 3
Description	Generalized Joint Hypermobility (GJH) Beighton Score: Prepubertal children and adolescents > 6 Men and women, post-puberty up to age 50 > 5 Men and women older than 50 > 4. If the Beighton score is 1 point below the cutoff and the 5PQ* is "positive" (at least 2 positive items), a diagnosis of GJH may be made.	2 or more of the following features (A-C) must be present (i.e. A and B, A and C, B and C, or A and B and C): A. Systemic manifestations of a more generalized connective tissue disorder (a total of five must be present): 1. Unusually soft or velvety skin 2. Mild skin hyperextensibility 3. Unexplained striae such as striae distensae or rubrae at the back, groins, thighs, breasts and/or abdomen in adolescents, men or prepubertal women without a history of significant gain or loss of body fat or weight 4. Bilateral piezogenic papules of the heel 5. Recurrent or multiple abdominal hernia(s) (e.g., umbilical, inguinal, crural) 6. Atrophic scarring involving at least two sites and without the formation of truly papyraceous and/or hemosideric scars as seen in classical EDS 7. Pelvic floor, rectal, and/or uterine prolapse in children, men or nulliparous women without a history of morbid obesity or other known predisposing medical condition 8. Dental crowding and high or narrow palate 9. Arachnodactyly, as defined in one or more of the following: (i) positive wrist sign (Steinberg sign) on both sides; (ii) positive thumb sign (Walker sign) on both sides 10. Arm span-to-height ≥ 1.05 11. Mitral valve prolapse (MVP) mild or greater based on strict echocardiographic criteria 12. Aortic root dilatation with Z-score > +2 B. Positive family history C. Musculoskeletal complications (must have at least one): 1. Musculoskeletal pain in two or more limbs, recurring daily for at least 3 months 2. Chronic, widespread pain for ≥ 3 months 3. Recurrent joint dislocations or frank joint instability, in the absence of trauma (a or b)	All prerequisites required: 1. Absence of unusual skin fragility, which should prompt consideration of other types of EDS 2. Exclusion of other heritable and acquired connective tissue disorders, including autoimmune rheumatologic conditions 3. In patients with an acquired /autoimmune connective tissue disorder additional diagnosis of hEDS requires meeting both Features A and B of Criterion 2. Feature C of Criterion 2 (chronic pain and/or instability) cannot be counted towards a diagnosis of hEDS 4. Exclusion of alternative diagnoses that may also include joint hypermobility by means of hypotonia and/or connective tissue laxity

	a. Three or more atraumatic dislocations in the same joint or two or more atraumatic dislocations in two different joints occurring at different times b. Medical confirmation of joint instability at two or more sites not related to trauma

* The Five-Point Questionnaire, adapted from Grahame and Hakim, 2003

Criterion 1, 2, and 3 quoted from Malfait et al. (2017, p 16-18).

Table 3. Clinical Classification of the Ehlers-Danlos Syndromes, Inheritance Pattern, and Genetic Basis (Malfait et al., 2017)

	Clinical EDS subtype	Abbreviation	IP	Genetic Basis	Protein
1	Classical EDS	cEDS	AD	Major: *COL5A1, COL5A1*	Type V collagen
				Rare: *COL1A1*	Type I collagen
				c.934C>T, p.(Arg312Cys)	
2	Classical-like EDS	clEDS	AR	*TNXB*	Tenascin XB
3	Cardiac-valvular	cvEDS	AR	*COL1A2* (biallelic mutations that lead to *COL1A2* NMD and absence of pro α2(I) collagen chains)	Type I collagen
4	Vascular EDS	vEDS	AD	Major: *COL3A1*	Type III collagen
				Rare: *COL1A1*	Type I collagen
				c.934C>T, p.(Arg312Cys)	
				c.1720C>T, p.(Arg574Cys)	
				c.3227C>T, p.(Arg1093Cys)	
5	Hypermobile EDS	hEDS	AD	unknown	unknown
6	Arthrochalasia EDS	aEDS	AD	*COL1A1, COL1A2*	Type I collagen
7	Dermatosparaxis EDS	dEDS	AR	*ADAMTS2*	ADAMTS-2
8	Kyphoscoliotic EDS	kEDS	AR	*PLOD1*	LH1
				FKBP14	FKBP22
9	Brittle Cornea syndrome	BCS	AR	*ZNF469*	ZNF469
				PRDM5	PRDM5
10	Spondylodysplastic EDS	spEDS	AR	*B4GALT7*	β4GalT7
				B3GALT6	β3GalT6
				SLC39A13	ZIP13
11	Musculocontractural EDS	mcEDS	AR	*CHST14*	D4ST1
				DSE	DSE
12	Myopathic EDS	mEDS	AD or AR	*COL12A1*	Type XII collagen
13	Periodontal EDS	pEDS	AD	*C1R*	C1r
				C1S	C1s

IP, inheritance pattern; AD, autosomal dominant; AR, autosomal recessive, NMD, nonsense-mediated mRNA decay

University of Chester

UNIVERSITY OF CHESTER, DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL

WORKING TITLE: *The impact of external support in helping people with Ehlers-Danlos Syndrome live with the condition*

A. Applicant & Personnel

Applicant: *Kate Appleby*
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Applicant status: Staff ☐ Postgraduate Research ☐ Postgraduate Taught X Undergraduate ☐ Module Number: *PS7112*

Supervisor, if applicant is a student: *Dr Janine Carroll* Email: *j.carroll@chester.ac.uk*

Additional personnel 1: *Click here to enter text.* Email: *Click here to enter text.*
Role: *Click here to enter text.* Tel: *Click here to enter text.*
Additional personnel 2: *Click here to enter text.* Email: *Click here to enter text.*
Role: *Click here to enter text.* Tel: *Click here to enter text.*
Attach details of any additional personnel

B. SUBMISSION TYPE

1. What is the submission type?

X **First submission to this or any other committee**
☐**Resubmission of a rejected application by this committee** → Attach previous submission
↳ Summarise the changes made to the application since it was last considered by this committee, with reference to the committee's comments: *Click here to enter text.*
☐**First submission to this committee; has been submitted to another committee.**
↳Give details of the previous submission. Include committee name, date of submission and outcome. *Click here to enter text.* → Attach previous submission → Go to Section C
☐**Revised submission intended to replace an application approved by this committee**
↳ Give details of the previous submission date and any changes that have been made. → Attach previous submission
Click here to enter text.

C. FUNDING

2. Is the project subject to external funding?

X No → Go to Section D
☐Yes → Is funding secured? ☐No → Provide details: *Click here to enter text.*
☐Yes Funding body and mailing address: *Click here to enter text.*
Grant number, if applicable: *Click here to enter text.* Named PI: *Click here to enter text.*

D. NATURE OF RESEARCH

3. Are you a member of staff applying for approval for a student related research exercise?

X No → Go to Section E ☐Yes → Module code and name *Click here to enter text.*

i. Will the student/s be collecting data unsupervised and outside of lecture/lab time ☐No → Go to Section E
☐Yes→ Provide details of how ethical standards will be maintained Attach necessary documentation. *Click here to enter text.*

E. RESEARCH PLAN & METHODOLOGY

Provide a detailed description of the proposed research. You should expect to write a paragraph on each section. Please note that there is a requirement to show that the project is well formulated in terms of drawing on relevant literature and is methodologically, analytically and scientifically sound.

4. **Rationale/background** (theoretical justification for conducting the research): *Ehlers Danlos Syndrome (EDS) is a heritable connective tissue disorder, with several variants; the most common being classic type EDS, and hypermobility type EDS. Symptoms can include: stretchy and/or fragile skin, which bruises easily, and heals poorly; hyper-flexible, unstable joints which may be prone to frequent dislocation; myalgia and arthralgia; cardiovascular and gastric complications are not uncommon. Anecdotally individuals with EDS report poor understanding of EDS by medical professionals (both the nature of the condition itself and that it has commonly associated conditions such as IBS, Chronic Pain, Fatigue, Fibromyalgia, POTs, Arnold-Chiari malformation, pregnancy complications, etc) and that the support available is inconsistent. Misdiagnosis is common and it has been reported that the average length of time from onset of symptoms to diagnosis is 17 years. As EDS is often hard to observe externally, this condition can be difficult to diagnose and may be difficult for people without EDS to understand. There is no cure for EDS; treatment options are palliative, and may include; physiotherapy, occupational therapy, pain management, and assistive devices or adaptations, (such as wheelchairs, braces, crutches, hand rails, etc) as well as symptomatic treatment of injuries and related conditions. EDS is associated with poorer than average health-related quality of life and higher that average levels of anxiety and depression. This study is intended to help fill a gap in the research; there are studies on other chronic illnesses and the role of support, however little regarding EDS and the impact of external support in helping people with EDS live with their condition. Much of the existing research concerning the lived experience of EDS patients is in the context of the North American healthcare system; I am interested in how people located in the UK, specifically in the North of England perceive, and experience, support.*

5. **Aims and objectives** (expected and desired outcomes of the research; expected impact of the research): *Typically, people with EDS in the UK are treated under the NHS, which is a world-leader in the provision of evidence-based care. This study intends to explore the role of support (both NHS-provided, and social support) in improving the quality of life for people with this life-long chronic condition. This study could pave the way for future research which could inform the provision of services for people with EDS; for example utilising a quality of life measure such as EQ-5D to enable a stratification analysis of themes associated with the highest and lowest scoring participants, or validating a perceived mattering scale to allow evaluation of the role of perceived mattering in participants perception of support.*

6. **Research questions/hypotheses** (what you expect to learn): *What is the impact of external support in helping people with Ehlers-Danlos Syndrome live with the condition?*

7. **Procedure** (provide a summary of how you will conduct the research. More detailed responses should be given in the appropriate sections of the form, you may refer to them here): *Once ethical approval has been granted, permission to recruit participants from appropriate support groups (both online and face-to-face) will be sought. Having gained permission, recruitment messages will be posted to several Facebook groups, posters will be displayed on UoC parkgate campus, and I will attend face-to-face support groups to deliver recruitment information. When participants are identified, interview sessions will be booked. At interview sessions, participants will be given information packs, including consent forms, and will be briefed on the study, their rights, and what will occur during the session. Interviews will be audio recorded. On completion of the interview, participants will be given debriefing information packs. Participants will have five working days after the interview to invoke their right to withdraw; after this, the interviews will be transcribed, and thematic analysis will be carried out on the text. Dr Janine Carroll will also analyse a transcript selection for analyst triangulation. The results will then be written up.*

8. **Proposed timetable for research** (include deadlines for data collection and contingency plans) : *23rd March: First submission to Ethics committee. 19th April: resubmission to Ethics committee (if required). May - June: Recruitment and data collection. May - July: Transcription. August: Analysis. August - September: Write-up completion. October: Submission deadline. My contingency plan is to run a similar study of members of the general public, rather than people with EDS, I would run that study to the same timetable. I would provide participants with a description of EDS, including descriptions of*

how EDS may affect a person's life, and interview them about what support means to them, what forms of support they think are available to people with this sort of condition, and what forms of support they think should be available. This would allow me to address similar topics: support; perceptions of support; and ideal support.

9. **Describe <u>any</u> risk of physical harm or psychological distress to participants, however minor, in the recruitment process, during data collection or post data collection**. Provide details of how you will minimise and manage any issues. <u>You must include details of your debrief procedures here</u>: *There are no risks of physical harm to participants posed by any stage of the study. I will be interviewing participants at a wheelchair-accessible public location/s, which will also facilitate access for participants who have mobility limitations, but who do not use wheelchairs, and thereby reduces risks of accidental physical injury at interview locations, such as stairs. There is a risk of psychological distress to participants should they be upset by discussing EDS-related topics. I will be recruiting participants diagnosed with EDS for more than 12 months; in part to reduce the risk of causing distress to participants by discussing a recent diagnosis. Potential participants will be advised that the study is recruiting adults (18+) who have been diagnosed with any variant of Ehlers-Danlos Syndrome (EDS), for over 12 months, and that they should not take part if they feel they may become upset by discussing the research topic. Interviews will be carried out only during office hours when Dr J Carroll is available to provide supervision should any unexpected situation occur during data collection. Interview questions are designed to query participants' perceptions of support, not their condition/s. Potential participants will be informed what to expect during the interview, and this focus of inquiry will be made explicit; by being clear that the interview will not be about the impact of EDS itself on participants' lives, it will be possible to steer interviews away from emotive, personal issues (see: appendix 1 - Participant Information Sheet). In addition, the final interview question is intended to provide positive mood induction (appendix 2 - interview questions). During debrief, participants will be given time to recover their composure, further chance to withdraw from the study, and will be provided a further information sheet, which includes contact details for relevant support services should they require them (SHS, HMSA, Samaritans, etc. See: appendix 3 - further information). Post data-collection: there is a risk of embarrassment or distress to participants should they be identified, so all interview transcripts will be anonymised. This will prevent exposure of information about participants' medical conditions or details of their experiences of support, which may not all be positive. If critical of friends, family, or healthcare professionals, exposure of participants' candid opinion may cause embarrassment, upset to vital relationships, or cause anxiety about future provision of healthcare.*

10. **Is there any deception involved in the study?**
    X No
    ☐Yes → Justify use of deception and provide debrief details: Click here to enter text.

F. SAMPLE SIZE, PARTICIPANTS AND RECRUITMENT

If you are utilising internet mediated data collection methods you must consult the relevant guidelines, consider them in this section and make your procedure clear, particularly for questions 20-24.

11. **Who do you intend to recruit for participation in your study?**
    ☐No recruitment
    ↳ ☐Pre-existing data ☐Media/online-media based research (eg: forums) ☐ Other → Explain: Click here to enter text.
    X Human participants
    ☐Non-human animal subjects OR Both non-human animal subjects and human participants
    ↳ If during the course of the research the costs to the individual animal/s rose above that expected, describe the point at which you would remove the animal from the research. Click here to enter text.
    ↳ Once the animal has been removed from the research describe how any distress and harm caused will be dealt with. Click here to enter text.
    ↳ If you are working with both human and non-human animal participants and during the course of the research the costs to the individual animal/s rose above that expected and were removed from the research is there any likely

18. Describe how your sample will be identified and how you obtained contact information. *Participants will be recruited from specialist support groups on Facebook (including but not limited to: "Ehlers-Danlos Initiatives, Survey and Unified Voices" https://www.facebook.com/groups/EhlersDanlosUnifiedVoices/1559716001015005/; "Woo-less EDS - an Ehlers-Danlos group for science" https://www.facebook.com/groups/987538107943310/; "Skeptical Spoonies - Evidence Based Chronic Pain Support" https://www.facebook.com/groups/387307311351023/; and "EDS - Zebras need Zebras" https://www.facebook.com/groups/262111370664209/), I shall also attend local support group meetings and (with permission) recruit participants from attendees, as well as contact by email or private message (Facebook) potential participants who are already known to me. Potential participants will be invited to contact me for further details via my UoC email address, and their contact details will be requested at that time. I will also display recruitment information in poster form at The UoC Parkgate campus.*

19. Indicate the types of recruitment to be used and attach copies of all materials. If you have not attached evidence explain why:
Pleases see appendices 5 and 6 for details of recruitment text and poster Check all that apply

Do you need permission to contact potential participants and/or display material?
☐No X Yes Explain and give details: *I will require permission to post recruitment details online, to contact potential participants directly via email (or equivalent), and will display the recruitment details in poster form on campus at The UoC Parkgate campus.*

☐I am using pre-existing/online/on-line media based data → Go to Section G
☐I am using non-human animal subjects and I have completed Q18. → Go to Section G
☐I am using human and non-human animal subjects and I have completed Q18 and provided information below.
☐ RPS Ensure you have the required number of credits
☐Letters/emails to potential participants
X Social media Ensure you have consulted BPS guidelines for internet mediated research and you must provide appropriate details in relevant sections.
X Flyers/posters/brochures
X Verbal script (face-to-face or telephone recruitment)
☐Websites
☐Powerpoint presentation
☐Newspaper/magazine advertisements ☐Radio/tv advertisements
☐Other *Click here to enter text.*

20. Indicate if this research exclude any persons from the participation or analysis stage on the basis of:
☐Gender ☐Ethnicity X Age ☐Sexual orientation ☐ Mental health issues ☐Specific learning difficulties
☐Physical factors (e.g. physical ability, visual acuity, language/accent, handedness etc)
☐Other *Click here to enter text.*

a) If you are excluding any participants on the basis of any of the above categories, please justify their exclusion and discuss how any issues of distress and/or embarrassment arising from the exclusion will be minimised, monitored and managed during this process. *Participants who are under 18 are being excluded as this study is intended to explore the impact of support on adults with EDS. Children who have EDS are provided different services to adults (e.g. NHS services), and it is expected that the types of support that children who have EDS typically require differs to the types of support that adults who have EDS typically require. Evaluating the role of support to children who have EDS would be a valid area of research, however would be better addressed by a separate study given children's differing needs and the types of services available to them*

No exclusions apply ☐ → Go to Q21

21. Will potential participants be asked any screening questions to determine whether they will be recruited?
☐No → Go to Q22

☐Patients/clients
☐People in custody
☐People engaged in illegal activities (e.g. drug-taking)
↳ If any of the above boxes are checked consult BPS guidelines on the protection of vulnerable persons. If you are a student, consult with your supervisor before continuing with your application.
X None of the above → Go to Q18

If you are working with vulnerable persons, ascertain whether it is necessary to obtain satisfactory DBS clearance (or equivalent for overseas students) for all applicants who will be in contact with vulnerable persons, then check one of the following.
☐DBS clearance obtained and shown to supervisor.
X DBS clearance is not necessary → Explain: *My participants do not fall under any of the above detailed categories, however, as people with chronic health conditions, which are likely to involve mobility issues, and who are from a population which is known to experience anxiety and depression at higher rates than the general population, I acknowledge that my participants can be considered to be derived from a vulnerable population. DBS clearance is not neccesary, however I do have DBS clearance to work with vulnerable persons, in a voluntary capacity, and the relevant documents have been made available to Dr Carroll, and attached to this application.*
Attach suitable documentary evidence. If you have not attached evidence explain why: *Please see appendix 4 - DBS certificate*

how EDS may affect a person's life, and interview them about what support means to them, what forms of support they think are available to people with this sort of condition, and what forms of support they think should be available. This would allow me to address similar topics: support; perceptions of support; and ideal support.

9. **Describe <u>any</u> risk of physical harm or psychological distress to participants, however minor, in the recruitment process, during data collection or post data collection**. Provide details of how you will minimise and manage any issues. <u>You must include details of your debrief procedures here</u>: *There are no risks of physical harm to participants posed by any stage of the study. I will be interviewing participants at a wheelchair-accessible public location/s, which will also facilitate access for participants who have mobility limitations, but who do not use wheelchairs, and thereby reduces risks of accidental physical injury at interview locations, such as stairs. There is a risk of psychological distress to participants should they be upset by discussing EDS-related topics. I will be recruiting participants diagnosed with EDS for more than 12 months; in part to reduce the risk of causing distress to participants by discussing a recent diagnosis. Potential participants will be advised that the study is recruiting adults (18+) who have been diagnosed with any variant of Ehlers-Danlos Syndrome (EDS), for over 12 months, and that they should not take part if they feel they may become upset by discussing the research topic. Interviews will be carried out only during office hours when Dr J Carroll is available to provide supervision should any unexpected situation occur during data collection. Interview questions are designed to query participants' perceptions of support, not their condition/s. Potential participants will be informed what to expect during the interview, and this focus of inquiry will be made explicit; by being clear that the interview will not be about the impact of EDS itself on participants' lives, it will be possible to steer interviews away from emotive, personal issues (see: appendix 1 - Participant Information Sheet). In addition, the final interview question is intended to provide positive mood induction (appendix 2 - interview questions). During debrief, participants will be given time to recover their composure, further chance to withdraw from the study, and will be provided a further information sheet, which includes contact details for relevant support services should they require them (SHS, HMSA, Samaritans, etc. See: appendix 3 - further information). Post data-collection: there is a risk of embarrassment or distress to participants should they be identified, so all interview transcripts will be anonymised. This will prevent exposure of information about participants' medical conditions or details of their experiences of support, which may not all be positive. If critical of friends, family, or healthcare professionals, exposure of participants' candid opinion may cause embarrassment, upset to vital relationships, or cause anxiety about future provision of healthcare.*

10. **Is there any deception involved in the study?**
    X No
    ☐Yes → Justify use of deception and provide debrief details: Click here to enter text.

F. SAMPLE SIZE, PARTICIPANTS AND RECRUITMENT

If you are utilising internet mediated data collection methods you must consult the relevant guidelines, consider them in this section and make your procedure clear, particularly for questions 20-24.

11. **Who do you intend to recruit for participation in your study?**
    ☐No recruitment
    ↳ ☐Pre-existing data ☐Media/online-media based research (eg: forums) ☐ Other → Explain: Click here to enter text.
    X Human participants
    ☐Non-human animal subjects OR Both non-human animal subjects and human participants
    ↳ If during the course of the research the costs to the individual animal/s rose above that expected, describe the point at which you would remove the animal from the research. Click here to enter text.
    ↳ Once the animal has been removed from the research describe how any distress and harm caused will be dealt with. Click here to enter text.
    ↳ If you are working with both human and non-human animal participants and during the course of the research the costs to the individual animal/s rose above that expected and were removed from the research is there any likely

distress caused to the human participant? Explain and give details of how you will minimise harm and distress: Click here to enter text.

☐Combination of the following: Check all that apply

↳ ☐Pre-existing data ☐Media/online-media based ☐ Other → Explain: Click here to enter text.

☐Human participants ☐Non-human animal subjects OR Both non-human animal subjects and human participants

12. What is the sample size for your study? (If you are a UG or PGT student you should discuss this with your supervisor.. If you are using pre-existing data or online/media based research, give details of the type and size of sample eg: number of participants; number, type and extract length of interviews/case studies/articles/programmes/films). *Up to 12 participants*

13. Was a statistical/power analysis conducted to determine the adequate sample?

☐ Yes → give details Click here to enter text.

X No → describe how you determined the sample size (where appropriate you should refer to Section E) *On advice from dissertation superviser, and after considering how much interview data can be meaningfully processed within the timeframe.*

14. Where will the proposed recruitment and/or data collection take place? (If you are using pre-existing data/on line/media based research you should still indicate a location and consider related health and safety issues and issues of data protection and storage in relevant sections of this form). Check all that apply

X A University of Chester campus → Give details: *Recruitment: I intend to display recruitment posters at Parkgate campus; once ethical approval for the study is granted I will consult UoC staff as to which locations on campus are suitable, and if permission is required, who I will need to obtain that from. (see: appendix x - recruitment poster). Data Collection: As a contingency plan - should a participant be unable to attend Altrincham library, or should that location become unavailable, I will seek permission to use a private room at Parkgate campus for data collection.*

☐Online (including RPS) → Before you continue, consult BPS guidelines for internet mediated research and you must provide appropriate details in relevant sections. E.g. participant information; informed consent; withdrawal procedures etc.

X Other site(s) → Give details: *Data Collection location: Staff at Altrincham Library have kindly given provisional permission to book a meeting room for interviews. The site is accessible to wheelchair users, close to wheelchair-accessible public transport links, and available free-of-charge. By utilising the public library, I will have adequate privacy for interviews (private room), whilst also being within hearing distance of library staff should an unexpected situation arise. Trafford council are responsible for the health and safety of people using Altrincham library.*

15. Have health and safety issues been adequately considered? Click here to enter text.

☐I am a UG or PGT student using pre-existing data and I have attended the recommended health and safety briefing.

☐Yes → *Office use only:* Confirmation of attendance Y ☐ N ☐

☐No → Explain why & provide details of alternative arrangements & considerations Click here to enter text.

X I am a UG or PGT student collecting data from human participants and/or non-human animal subjects and I have attended the recommended Health and Safety briefing.

X Yes → Confirmation of attendance Y ☐ N ☐

☐No → Explain why and provide details of alternative arrangements and considerations Click here to enter text.

☐I am a member of staff/PGR student and I have attached a risk assessment form. Attach suitable documentary evidence of permission. If you have not attached documentary evidence explain why. Click here to enter text.

16. Is permission to recruit potential participants/subjects required from an organisation other than the University of Chester?

X Yes → Explain: *I will approach the HMSA about the possibility of disseminating recruitment information at local meetings, and will request permission from online (facebook) group administrators prior to posting recruitment notices in their groups.* Attach suitable documentary evidence of permission. If you have not attached documentary evidence explain why. *I have not yet approached these organisations for permission as I wish to have received ethical approval for the focus of my study prior to this; should I impliment my alternative ('back-up') study plans, I may still be recruiting with the help of these groups, and do not wish to alter the focus of the study after having approached them for permission.*

☐No → Explain: Click here to enter text.

17. Will participants fall into any of the following special groups?

☐Schoolchildren (under 16 yrs of age)

☐People with learning or communication difficulties

X Yes → Explain and describe how you will minimise, monitor and manage any issues of distress and embarrassment: *Potential participants will be provided with the Participant Information Sheet (PIS), which states that the study is seeking participants who were diagnosed more than one year prior to their participation in the study. This is in part because the study is concerned with experiences and perceptions of support available to people with EDS, and so participants must have been living with the diagnosis for enough time to have experienced the full range of support available to them, several of which may only have become available on formal diagnosis. However, this exclusion is also in place to reduce the possibility of participant distress occurring as a result of discussing issues raised by potentially complex medical condition/s, typically forcing profound lifestyle adjustments. In addition, the PIS suggests that people who may be distressed by discussion of support or EDS should not take part. The PIS will be provided to potential participants on initial expression of interest, and before they arrange an interview session so that they are able to decide if the study is suitable for them to take part in, without pressure. The recruiters contact details are on this sheet and the prospective participants will be given the option to contact the researcher themselves, or give their contact details and be contacted. If they agree to give their contact details then no further contact will be made for 24 hours after potential participants receive a copy of the PIS. When potential participants are contacted it will be made explicit that they are volunteers and have no obligation to take part in the research. It will also be stipulated that the research will have no impact on the services they receive or may receive in future. If participants agree to give their contact details, they will only be contacted twice; once to reintroduce the research topic and to ask if the potential participant has had chance to read the PIS (or requires another copy); and one follow-up to ask if they are still interested in the research. At every point of contact the possible participant will be reminded that they are under no obligation to take part in the study, that all identifiable information will be anonymised, and that the recruiter understands that they may not have time or want to take part in the study. When consent for the study is sought during the interview sessions, participants will be asked to re-read the information and confirm they have read and understood it (appendix 7 – consent form). It is anticipated that people who have lived with such a diagnosis for more than a year, who are recruited through organisations (i.e. support groups and the UoC), and who choose to participate knowing the focus of the study, having had opportunity to not provide contact details and withdraw all interest, are less likely to be distressed by discussion of the topic. Should correspondence or conversation with potential participants suggest they are distressed or embarrassed, they will be offered support information as detailed in the debrief information (appendix 3); should it seem necessary, Dr Carroll would be advised and supervisory guidance sought.*

22. How will informed consent be sought? *The Participant Information Sheet outlines the purpose of the study; the anticipated outcome; their rights as study participants; the potential benefits and disadvantages to participating in such studies, and their right to withdraw will be emphasised. Participants will be provided with this before they arrange an interview session. At the start of the session, before the interview, they will be asked to read it again prior to reading and signing the consent form.*

23. How will anonymity and confidentiality be maintained during recruitment and data collection? *During recruitment, potential participants will be encouraged to initiate contact by email if they wish to take part; if I am contacted via Facebook private message I shall ask them to email my UoC email account for further details. If potential participants are happy to provide their email address, I will email them from my UoC account using the standard first contact email detailed in appendix 5 – recruitment text. Participants will be assigned pseudonyms, and interview text that identifies locations, participants or other persons will be redacted from the transcripts, to preserve anonymity. All participant-identifying data, such as the participant-pseudonym key and participant contact details will be stored in a password-protected file, which will be stored on password-protected devices; a PC, and backed up to an external drive, which will also be password protected as well as encrypted. Files will be transported only on a recording device until they can be transferred to a password protected, encrypted device. Once they are transferred, they will only be transported by password protected, encrypted portable drive.*

24. How will participants be able to withdraw from data collection? *Verbally at any time during the interview process, or by emailing myself or Dr J Carroll.*

↳ Is there a time limit for withdrawal? Explain: *The withdrawal deadline will be five working days after the interview; transcription will take place only after this deadline has passed.*

↳ What will happen to any partially collected data? Explain: *Partially completed interviews will be retained only if - the participant wishes to finish the session, for any reason, but does not wish to withdraw from the study, does not appear to be distressed, and gives audio recorded verbal consent to retention of the data for the transcript. Otherwise the data will be discarded.*

25. What is the time commitment expected of participants? *Approximately one hour, plus travel time.*

26. Indicate the type and amount of compensation participants will receive. X None

Amount value: *Click here to enter text.*. ☐Money: ☐Gift certificate: ☐Travel Expenses: ☐Other: Explain: *Click here to enter text.*

27. Indicate where the following information will be available to participants. Attach documentary evidence. Check all that apply.

	Information. sheet	Letter	Email	Email info. page	Consent Form	PowerPoint	N/A
Brief details about the purpose of the study	X	☐	☐	☐	X	☐	☐
Contact details for further information	X	☐	X	☐	☐	☐	☐
Explanation of how and why participant has been chosen	X	☐	☐	☐	☐	☐	☐
Notification that materials/interviews are not diagnostic tools/therapy or used for staff review/development purposes	X	☐	☐	☐	☐	☐	☐
Explanation participation is voluntary	X	☐	☐	☐	☐	☐	☐
Details of any incentives or compensation	☐	☐	☐	☐	☐	☐	X
Details of how consent will be obtained	X	☐	☐	☐	X	☐	☐
If research is observational, consent to being observed	☐	☐	☐	☐	☐	☐	X
Details of procedure so participants are informed about what to expect	X	☐	☐	☐	☐	☐	☐
Details of time commitments expected	X	☐	X	☐	☐	☐	☐
Details of any stimuli used	☐	☐	☐	☐	☐	☐	X
Explanation of right to withdraw and right to withdraw procedure	X	☐	☐	☐	☐	☐	☐
Option for omitting questions participant does not wish to answer	X	☐	☐	☐	☐	☐	☐
Procedure regarding partially completed questionnaires or interviews	X	☐	☐	☐	☐	☐	☐
With interviews, information regarding time limit for withdrawal	X	☐	☐	☐	X	☐	☐
Details of any advantages and benefits of taking part	X	☐	☐	☐	☐	☐	☐
Details of any disadvantages and risks of taking part	X	☐	☐	☐	☐	☐	☐
Information that data will be treated with full confidentiality and that, if published, those data will not be identifiable as theirs	X	☐	☐	☐	X	☐	☐
Debriefing details	X	☐	☐	☐	☐	☐	☐
Dissemination information	X	☐	☐	☐	☐	☐	☐
Further information (relevant literature; support networks etc)	X	☐	☐	☐	☐	☐	☐

If you have checked n/a for any of the above, provide further details: *There are no incentives or compensation available for participation in this study. This study does not involve observation of participants. This study does not utilise stimuli.*

Ensure you have provided further details regarding the above in the relevant sections of the form and attached any necessary documentation. If you have not attached the necessary documentation explain why:

G. DATA COLLECTION

28. Indicate the types of data collection methods that will be used Attach copies of all materials (where appropriate

Check all that apply

☐I am using pre-existing data and have indicated all the original methods of data collection below.
☐Online/online-media based research answer 28(a) and (b)
☐Observations Diaries/Journals completed by researcher attach any instructions given to participants and any multi-media stimuli answer 28(a) and (b)
☐Observations Diaries/Journals completed by participants attach any instructions given to participant and any multi-media stimuli answer 28(a) and (b)
☐ Questionnaires/Surveys attach version of questionnaire that will be used in study, answer 28(a) and (b)
X Individual interviews attach list of questions/topics and any multi-media stimuli, answer 28(a) and (b)
☐Focus groups attach list of questions/topics and any multi-media stimuli, answer 28(a) and (b)
☐Biological specimens (e.g. blood, urine) Go to Q29
☐Biomedical devices (e.g. Biopac) Go to Q29
☐Cognitive measures (e.g. Reaction time, accuracy, recognition) attach copies of stimuli and answer 28 (a) and (b
☐Multimedia stimuli attach original material (where appropriate) URL links or other relevant information and answer 28 (a) and (b)
↳ ☐Video/DVD
☐Online/video gaming footage
☐Web sites/On-line forums
☐Written text (e.g. newspapers, magazine, books, transcriptions, scenarios, vignettes)
☐Audio (e.g. radio broadcasts, recordings)
☐Still images
☐Stimuli made from recordings of other persons that are not in the public domain (e.g. personal photographs, video/audio recordings)
↳ If stimuli are identifiable, obtain consent for their use attach evidence of consent

a) Does the content of the material contain anything that could cause distress or alarm and/or involve sex, violence, substance abuse, profanity, impudence or other types of mature content? Fully consider the suitability of the stimuli and the possible impact on the participant/researcher attach original material (where appropriate) URL links or other relevant information
XNo → Briefly describe the content: *No multi media (or other) stimuli; interview questions attached*
☐Yes
↳ Is the material from a source that has been given a universally acceptable certification OR has the source been considered by an appropriate agency with regard to suitability for audiences in terms of its ability to cause distress or alarm and/or in terms of content issues involving sex, violence, substance abuse, profanity, impudence and other types of mature content? (eg: material used by multi national media organisations and widely accessible by general audiences)
☐Yes → Provide details and justify the use of the material. Explain how your will minimise, monitor and manage any issues of distress to the participant and/or researcher Click here to enter text.
☐No/not sure → Explain, provide details and justify the use of the material. Explain how you will minimise, monitor and manage any issues of distress to the participant and/or researcher.

b) Once data collection is complete what action will be taken to ensure that participants and/or researchers leave the research in a positive frame of mind? *Final interview question is intended as a positive mood inducement*

29. How will you collect your data? Check all that apply

☐ I am conducting an experiment Provide full procedural details Click here to enter text.
☐ I am using observations/diaries/journals Provide full procedural details Click here to enter text.
X I am conducting surveys/interviews/focus groups Provide full procedural details *I will collect my data by conducting face-to-face interviews. The interviews will be recorded by digital audio device, and then transcribed for analysis. At interview sessions, participants will be given information packs, including consent forms, and will be briefed on the study, their rights, and what will occur during the session. On completion of the interview, participants will be given debriefing information packs. Participants will have five working days after the interview to invoke their right to withdraw; after this, the interviews will be transcribed, and thematic analysis will be carried out on the text. Dr Janine Carroll will also analyse a transcript selection for analyst triangulation.*

☐ I am conducting internet based research Provide full procedural details Click here to enter text.

☐ I am conducting media based research Provide full procedural details Click here to enter text.

☐ I am using pre-existing data. Provide full details of how the data was originally collected making specific reference to key ethical considerations of management of harm & distress, consent, anonymity & confidentiality Click here to enter text.

30. Will you make any recordings of human participants? (interview/focus groups, observations, images of participants' bodies)

X Yes→ Go to question 31 ☐No → Go to section H

31. What will be recorded? Check all that apply

X Interview ☐Focus Group ☐Images of participants' bodies ☐Observations ☐Other → Explain: *Click here to enter text.*

32. How will the data be recorded? Check all that apply

☐Video X Audio ☐Photographs ☐Written transcripts ☐Other → Explain: *Click here to enter text.*

33. Can participants' identities be determined from the recording? (If the recording is a facial photograph/video or audio recording of a voice, the correct answer is 'yes'.)

☐ No

X Yes → Describe how you will protect privacy and anonymity during transcription and analysis. *Interviews will be transcribed with identifying details obscured, using pseudonyms to protect participants privacy and anonymity. I will be carrying out all transcription; no other person will have access to data or records that could identify individuals, except for academic staff.*

34. Will the recordings be destroyed? NOTE: Participants must consent to their recordings being retained/archived.

X Yes → Explain how and when *The recordings will be retained for the minimum time required by UoC regulations; all copies (including back-ups) will be permanently deleted, and only anonymised transcripts will be retained, as needed.*

☐ No→ Justify retaining the recordings and attach evidence of consent: *Click here to enter text.*

35. Will the recordings be used outside of this research study? NOTE: Participants must consent to all outside uses of their recordings.

X Yes → Answer Question 36 and attach evidence of consent

☐ No → Go to Section H

36. How will the recordings be used outside of this research study? Check all that apply

☐ Shared with other researchers not listed on this application: Explain: *Click here to enter text.*

X Used for research dissemination (conferences, journals, media publications, consultancy) *I intend to seek publication of the results of this study after completion of the module; I have discussed the possibility of submitting this to a relevant journal with Dr Carroll.*

☐ Used for educational purposes (e.g. training, teaching): Explain: *Click here to enter text.*

☐ Used within a commercial/public organisation: Explain: *Click here to enter text.*

☐ Other → Explain: *Click here to enter text.*

37. When the recordings are used outside of this research study, will they contain identifiable information (e.g. names, facial photographs/video, unmodified voices)?

☐ No

X Yes

↳ Will disclosure of participants' identity outside this research study reasonably place participants at risk for criminal or civic liability or be damaging to participants' financial standing, employability or reputation?

☐ Yes → Explain why it is necessary to disclose participants' identity: *Click here to enter text.*

X No → Go to Section H

H. DATA ANALYSIS

38. Describe your methods of data analysis: *I will be using Thematic Analysis to identify themes in the interview text. Dr Carroll, or another academic staff member, will recode a selection of data collected.*

I. DATA PROTECTION AND STORAGE

39. Where and in what form will the research materials be stored? Describe fully the storage process during collection, analysis and archiving and consider issues of security.: *Participant data will be recorded and stored in a password protected file, on a password protected PC that is secured by appropriate up-to-date anti-malware software. Audio recordings will be made using a portable digital device which will be kept on my person until the file can be transferred to the PC and deleted from the recording device. All data will be backed up to an external password protected drive; should it be necessary to transport files to a different location, this will be done using a password protected portable drive, and accessed on UoC computers only; unless advised otherwise by academic staff – e.g. for submission to an external examiner.*

40. Will the research materials be destroyed on completion of the project?

☐Yes → Explain how and when:
X No → Explain why the materials need to be maintained: *Research materials will be maintained only as required by UoC regulations and in order to seek journal publication.*

41. Will the research materials include any identifying information (e.g. names, telephone numbers)?

X No
☐Yes → Describe the type of information and justify why it will be retained: *Click here to enter text.*
↳ Will the identifying information be deleted?
☐Yes → State when and justify the retention of the information until this time: *Click here to enter text.*
☐ No → Justify the retention of the information: *Click here to enter text.*

J. DISSEMINATION

42. How will the research results be shared?

X Academic assessment (e.g. dissertation; assignment report) Explain and give details: *This study is my dissertation project*
X Academic dissemination (e.g. Journal publication; conferences) (If you are an UG or PGT student you must discuss this with your supervisor before checking this box). Explain and give details: *I intend to seek journal publication of the results of this study; to be further discussed with my supervisor on completion of the study.*
☐Non-Academic dissemination (e.g. printed/online article) (If you are an UG or PGT student you must discuss this with your supervisor before checking this box). Explain and give details: *Click here to enter text.*
☐Academic learning & teaching (e.g. class based research exercise) Explain and give details: *Click here to enter text.*
☐Consultancy (If you are an UG or PGT student you must discuss this with your supervisor before checking this box). Explain and give details: *Click here to enter text.*
☐Commercial/public sector. (If you are an UG or PGT student you must discuss this with your supervisor before checking this box). Explain and give details: *Click here to enter text.*
X Other (If you are an UG or PGT student you must discuss this with your supervisor before checking this box). Explain and give details: *Participants will have the option to request a one-page summary of my findings on completion of the study*

43. How will privacy and confidentiality be maintained during dissemination? *Participants will be assigned pseudonyms, and potentially identifying information, such as location details or names of other people referred to, will be redacted from*

the transcriptions. Participants will be referred to by pseudonym only during analysis, reporting and dissemination of the study; as personal details will be redacted prior to data analysis, they will not be detailed in any reporting of the study.

44. **Are there any specific considerations about sharing the research?** (eg: Is the data from friends and family and potentially embarrassing/upsetting for someone who reads it? Is the data relating to employee satisfaction/wellbeing and likely to be seen by senior staff?). *Study participants may include friends and acquaintances, and interviews may involve reference to personal medical information such as diagnosis of conditions other than EDS, or details of prior distress or mental health issues. This could be potentially embarrassing for interviewees if read by someone who could identify them. As well as redacting personal details from the transcripts, and reporting comments made anonymously, participants will be encouraged to refrain from disclosing such details if they are not comfortable doing so, should the course of the interview prompt it.*

University of Chester

UNIVERSITY OF CHESTER DEPARTMENT OF PSYCHOLOGY
APPLICATION TO DEPARTMENTAL ETHICS COMMITTEE

YOU HAVE NOW COMPLETED THE APPLICATION FORM. PLEASE READ AND SIGN THE FOLLOWING DECLARATION:

I confirm that I have familiarised myself with the regulatory codes and codes of conduct and ethics relevant to my area of research, including those of relevant professional organisations and ensure that the research which I propose is designed to comply with such codes.
I have familiarised myself with the following:
Department of Psychology Ethical Approval for Research: Procedural Guidelines.
University of Chester Research Governance Handbook
BPS Code of Ethics
BPS Code of Human Research Ethics
BPS Guidelines for Internet-mediated Research
BPS Research Guidelines and Policy Documents

I confirm I understand that:
All applications must be submitted according to the guidelines set out, assessed by at least 2 reviewers and are subject to discussion by departmental ethics committee. Data collection is not permitted until applications have been approved.
Collecting data without ethical approval is a serious breach of the BPS Code of Ethics.
Any change of plans to the research after the approval MUST be discussed by ethics committee. chair's action may be taken for minor changes.

Print the completed form off onto BLUE paper with the appendices on white paper. Handwritten applications are not accepted. Submit to the department office by the agreed deadline. Applications submitted after this deadline will not be processed until the following committee meeting.
If you are a member of staff or a PGR student, in addition to 2 paper copies you MUST submit an electronic version to c.leach@chester.ac.uk.

DATE: 16/03/2016

PRINT NAME: Kate Appleby

SIGNATURE: Kate Appleby

NOTES ON THE ROLE AND FUNCTION OF THE DEPARTMENT OF PSYCHOLOGY ETHICS COMMITTEE.

- *All decisions of the committee are based on the application form and reviewers comments ONLY. Forms should be as detailed and clear as possible. Verbal discussions are not considered as part of the application or review process.*
- *The review process strictly adheres to the University of Chester Research Governance Handbook and the BPS Code of Ethics.*
- *The decision of the committee is final. If you are a UG, PGT or PGR student you should discuss the decision of the committee with your supervisor. If you are a member of staff you may contact the chair of the committee for further clarification.*

ETHICS COMMITTEE DATE : Click here to enter a date.

CHAIRS COMMENTS: *Click here to enter text.*

Copies of letters granting access will need to be prondea ,

Consider phone / skype interviews, act on and amend with respect to reviewers Comments.

☐ **ACCEPTABLE**

Action: You may now commence with data collection subject to approval from any relevant external agencies.

DATA COLLECTION IS NOT PERMISSABLE UNDER THESE CONDITIONS

☑ **ACCEPTABLE SUBJECT TO SUBMISSION OF AMENDMENT FORM**

☑ **Acceptable subject to conditions listed by chair. Discuss conditions highlighted with supervisor and submit ethics application amendment form direct to office.**

☐ **Acceptable subject to conditions listed by chair: Submit ethics application amendment form direct to office.**

☐ **ACCEPTABLE SUBJECT TO CONDITIONS LISTED BY CHAIR:**

Action: Resubmit application for full review ensuring you have completed section B

☐ **REVISE AND RESUBMIT:**

Action: Resubmit application for full review ensuring you have completed section B

SIGNATURE: Moira Chappell

Office Use Only ***DOPEC NUMBER***

Appendices:

Appendix 1: Participant Information Sheet

Appendix 2: Interview questions

Appendix 3: Further information (debrief package)

Appendix 4: DBS certificate

Appendix 5: Recruitment text

Appendix 6: Recruitment poster

Appendix 7: Consent form

Appendix 1: Participant Information Sheet

Evaluating the role of external support in helping people with EDS live with their condition

Participant Information Sheet

You are being invited to participate in a research study. Before you decide whether to participate, it is important for you to understand why the research is being done and what it will involve. Please take the time to read the following information carefully and feel free to ask the interviewer if you would like any further information, or if there is anything you do not understand. I would like to stress that you do not have to accept this invitation, and should only take part if you want to.

Thank you for reading this.

What is the purpose of this study?

This study is evaluating how support can help people with Ehlers-Danlos Syndrome (EDS) manage to live with their condition, and to ask what your experiences of different types of support have been.

What will I be asked to do?

After having read this information sheet you will be asked to sign a consent form, and then we will begin the interview. If you have not participated in a research study of this kind before, this interview may seem quite different to other interviews you may have had in the past; it is not a diagnostic tool or therapy, nor is it intended for development purposes. There is no set time for your answers so you can answer in as much detail as you wish to. The interviews are expected to last up to an hour. The questions are about what you understand by the term 'support', and how you have experienced support. The interview will be recorded and transcribed for use in this study. Your name and any potentially identifying information (such as location or the names of people you may talk about) will be removed from the transcript, so you will not be identifiable as a study participant. When the interview is complete you will be asked to read an information sheet containing further information.

Who is conducting this study?

As part of the dissertation module, Kate Appleby, a postgraduate student on the MSc Psychology (Conversion) course at the University of Chester will be conducting and transcribing the interviews.

Is participation voluntary?

Participation in the study is entirely voluntary and you will not receive any formal compensation or reward (financial or otherwise) for your time. You can withdraw from the study at any time before, or during, the interview and any data that has been collected prior to your withdrawal will be destroyed. You do not have to provide a reason for withdrawing, and you can do so by speaking to the researcher during the interview, or alternatively by email. If you wish to withdraw after the interview, email Kate Appleby (1521754@chester.ac.uk), or my supervisor Dr Janine Carroll (email: j.carroll@chester.ac.uk) quoting your unique ID. You will have the right to withdraw from

this study until five working days after completion of your interview; after this time, the interview will be transcribed and you will not be able to withdraw as the interview will be part of the study analysis.

If for any reason you do not wish to complete the interview, or do not wish to answer a question, but do not wish to withdraw from the study, you can say so at any time during the interview. You will be asked to confirm that you consent to the retention of the interview recording and then to read an information sheet containing further information.

What are the risks/ benefits of taking part in the study?

Many people find taking part in research studies to be a satisfying experience and I hope this will be the same for you.

The interview will be about your experiences of support subsequent to your diagnosis of EDS; ***the researcher will not ask you about how EDS, or the diagnosis of EDS, has impacted your life***. This interview is not expected to cause any harm or distress, however ***if you find talking about EDS upsetting, or if you have experienced distressing or stressful evaluations (e.g. ATOS or healthcare professionals), or think talking about support may upset you, please do not take part in this study.***

Who can take part?

Any adult (18+) who has been diagnosed with any variant of Ehlers Danlos Syndrome (EDS), for over 12 months, may take part in this study.

What about the results?

The information collected will be analysed and although some comments made may be quoted in subsequent scientific publications, this will be done anonymously. Only myself as the researcher, my supervisor (Dr Janine Carroll, email: j.carroll@chester.ac.uk), and other academic staff such as an external examiner will have access to the transcript and audio recordings. If you would like to know more about the results of the study, you can give permission for us to retain your contact details for the purpose of sending you a copy. The results will be written in the form of a one page summary of what has been found.

What about confidentiality?

All interview recordings and your personal details are confidential, and will be kept secure by the researcher in password protected devices.

What if I have further questions?

If you have further questions, or any concerns about this study, please feel free to contact Kate Appleby (email: 1521754@chester.ac.uk) who will try to answer any queries you may have, alternatively you may contact my supervisor, Dr Janine Carroll (email: j.carroll@chester.ac.uk).

Ethical Approval

Ethical approval for this study has been sought and obtained from the University of Chester, Department of Psychology Ethics Committee.

Appendix 1: Participant Information Sheet

Evaluating the role of external support in helping people with EDS live with their condition

Participant Information Sheet

You are being invited to participate in a research study. Before you decide whether to participate, it is important for you to understand why the research is being done and what it will involve. Please take the time to read the following information carefully and feel free to ask the interviewer if you would like any further information, or if there is anything you do not understand. I would like to stress that you do not have to accept this invitation, and should only take part if you want to.

Thank you for reading this.

What is the purpose of this study?

This study is evaluating how support can help people with Ehlers-Danlos Syndrome (EDS) manage to live with their condition, and to ask what your experiences of different types of support have been.

What will I be asked to do?

After having read this information sheet you will be asked to sign a consent form, and then we will begin the interview. If you have not participated in a research study of this kind before, this interview may seem quite different to other interviews you may have had in the past; it is not a diagnostic tool or therapy, nor is it intended for development purposes. There is no set time for your answers so you can answer in as much detail as you wish to. The interviews are expected to last up to an hour. The questions are about what you understand by the term 'support', and how you have experienced support. The interview will be recorded and transcribed for use in this study. Your name and any potentially identifying information (such as location or the names of people you may talk about) will be removed from the transcript, so you will not be identifiable as a study participant. When the interview is complete you will be asked to read an information sheet containing further information.

Who is conducting this study?

As part of the dissertation module, Kate Appleby, a postgraduate student on the MSc Psychology (Conversion) course at the University of Chester will be conducting and transcribing the interviews.

Is participation voluntary?

Participation in the study is entirely voluntary and you will not receive any formal compensation or reward (financial or otherwise) for your time. You can withdraw from the study at any time before, or during, the interview and any data that has been collected prior to your withdrawal will be destroyed. You do not have to provide a reason for withdrawing, and you can do so by speaking to the researcher during the interview, or alternatively by email. If you wish to withdraw after the interview, email Kate Appleby (1521754@chester.ac.uk), or my supervisor Dr Janine Carroll (email: j.carroll@chester.ac.uk) quoting your unique ID. You will have the right to withdraw from

Appendix 2: Interview questions

Numbered questions will be asked; sub questions (a, b, c, etc) *may* be asked to prompt more detailed answers.

1. For the recording can you state your name... and for the transcript, could you please state your age, gender, and length of time since you were diagnosed with EDS?

2. What does the term "support" mean to you?
    a. What different forms of support can you think of?
    b. Can you detail any examples of institutional support, such as services provided by the NHS, the HMSA, or other organisations?
    c. Can you detail any examples of non-institutional, or social support?

3. What types of support have you received?
    a. Did that work out how you thought it would/should?
    b. Can you tell me a little more about that?
    c. What other forms of support have you received?/ Are there any other forms of support you have received?

4. What do you think are the main barriers, or facilitators, to people receiving support?
    a. What do you think would have made it possible/ easier to access that avenue of support (more effectively)? (if a barrier)
    b. What avenues of support would you suggest to someone who has just received a diagnosis of EDS?

5. In an ideal world, what does your dream support package for someone who has EDS look like?
    a. If you could have anything at all, to support you, what would it be?
    b. What do you think would be a barrier to achieving this for people with EDS?
    c. What would help make this possible?

6. What is your most enjoyable regular/ routine activity? What helps you to relax? E.g hobby, social activity, fiction genre, etc
    a. When are you planning on doing that next (or near substitute; eg what are you reading right now?)
    b. Alternative: What plans do you have for the rest of the day/ for the weekend?

Questions to be used if the interview is ended before all questions are completely answered:

If cut short due solely due to time constraints rather than because the participant wishes to withdraw:

1. Thank you for your time; can I confirm that you are happy for me to use your answers to the questions we got through for the study? Are you able to stay a few minutes longer to read this (debriefing) information sheet?

If cut short because of participant distress:

1. Thank you for your time; I think we should finish the interview here. Please read this (debriefing) information sheet, there are several avenues of support listed here, and I suggest that you make use of them if you have concerns that have been raised by what we have been speaking about today.
    a. What plans do you have for the rest of the day?
    b. How are you getting home?
    c. Will there be someone with you later?

If participant indicates they wish to invoke their right to withdraw from the study:

1. Thank you for your time; I would like to stress that you do not have to give a reason for withdrawing, and it is your right to do so. Please read this (debriefing) information sheet, and we'll conclude the session. Any data that has been collected will be destroyed; if you have any further concerns, please feel free to contact myself or my supervisor, Dr. Janine Carroll - our contact details are in the information sheets I have provided you with.

Appendix 3: Further information (debrief package) | ID:

Evaluating the role of external support in helping people with EDS live with their condition

Further Information for Participants

Thank you for participating in this research study. The interview is now finished, before we conclude the session, please take the time to read the following information carefully and feel free to ask the researcher if you have any further questions, or have any concerns.

What is the purpose of this study?

This study is evaluating how support can help people with Ehlers-Danlos Syndrome (EDS) manage their condition, and what participant's experiences of different types of support have been.

Many people find taking part in this kind of research to be a satisfying experience and I hope this has been your experience. Basic research of this type can be useful for the provision of evidence based care, and is essential for demonstrating the need for further research into, and specialised services for, people with EDS and other complex chronic conditions.

If you have any further questions or concerns about this study, or its purpose, please contact myself (Kate Appleby, email: 1521754@chester.ac.uk) or my supervisor (Dr. Janine Carroll, email: j.carroll@chester.ac.uk).

What happens next?

The audio recording of the interview will be transcribed, and any details that could make it possible to identify you will be removed. Although some comments made may be quoted in subsequent scientific publications, this will be done anonymously. The transcript will be analysed, along with the transcripts of interviews with other participants, and the results will be written up and reported.

At the start of this session you were asked to sign a consent form, which asked if you would like to receive a one page summary of the findings from this study; if you would like to change your answer to that question, please let the interviewer know now, as you will not be able to amend this choice once the session has finished.

What if I decided to withdraw from the study?

Participation in the study is entirely voluntary, and you can withdraw from the study at any time before or during the interview, and up to five working days afterwards. *You do not have to provide a reason for withdrawing*, and you may have done so by speaking to the researcher during the interview. If you wish to withdraw after the interview has finished, email Kate Appleby (1521754@chester.ac.uk), or my supervisor Dr Janine Carroll (email: j.carroll@chester.ac.uk) quoting your unique ID. After five working days the interview will be transcribed and you will not be able to withdraw as the interview will be part of the study analysis. If you decide to withdraw, any data recorded will be destroyed.

What if I have concerns about issues raised during the interview, such as the support available to me, or if I feel upset?

This interview is not expected to have caused harm or distress, however if you have found it to be upsetting, or troubling in any way, there are several avenues of support listed below:

Support for EDS:

Ehlers-Danlos Support UK: website: www.ehlers-danlos.org; helpline: 0800 907 8518 Monday, Tuesday, Wednesday and Friday (09:30 - 17:30)

HMSA (Hypermobility Syndromes Association): website: http://hypermobility.org; helpline: 03330 116 388

Support for mild to moderate Anxiety and Depression, or problems with sleep:

CBT programmes, such as 'Beating the Blues' or 'Sleepio' are free to access via the NHS; should you wish to access these short courses, ask your GP about how that can be arranged in your region. Many areas also offer other services such as crisis support, peer support, drop-in groups, and follow-up services for people who have previously accessed such courses.

In many areas of the North West of England these services can also be accessed via: **Self Help Services (part of the Big Life Group)**: website: www.selfhelpservices.org.uk; helpline: 0300 003 7029

Other support:

Samaritans are available 24 hours a day, 365 days a year -*"Just having someone to talk to that isn't family or friends can be a tremendous help. You do not need to be suicidal to get in touch."* Website: www.samaritans.org; helpline: 116 123

You can also contact your GP, or consultant, if you have concerns about any issues raised during the interview.

What if I have further questions?

If you have further questions, or any concerns about this study, please feel free to contact Kate Appleby (email: 1521754@chester.ac.uk) who will try to answer any queries you may have, alternatively you may contact my supervisor, Dr Janine Carroll (email: j.carroll@chester.ac.uk).

Thank you for participating in this study

Appendix 4: DBS certificate

STRICTLY PRIVATE AND CONFIDENTIAL

KATE APPLEBY
6 CROSSLEY ROAD
SALE
CHESHIRE

M33 6LG

81892301

Enhanced Certificate

Disclosure & Barring Service

Certificate Number 001415330061

Date of Issue: 19 AUGUST 2013

Applicant Personal Details

Surname: APPLEBY

Forename(s): KATE

Other Names: NONE DECLARED

Date of Birth: 20 FEBRUARY 1978

Place of Birth: HALIFAX

Gender: FEMALE

Employment Details

Position applied for:
ADULT WORKFORCE VOLUNTEER SUPPORT WORKER

Name of Employer:
THE BIG LIFE COMPANY

Countersignatory Details

Registered Person/Body:
UCHECK

Countersignatory:
ROSY CLARK

Police Records of Convictions, Cautions, Reprimands and Warnings

NONE RECORDED

Information from the list held under Section 142 of the Education Act 2002

NOT REQUESTED

Enhanced Certificate

Disclosure & Barring Service

Certificate Number 001415330061

Date of Issue: 19 AUGUST 2013

Applicant Personal Details

Surname: APPLEBY

Forename(s): KATE

Other Names: NONE DECLARED

Date of Birth: 20 FEBRUARY 1978

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Name of Employer:
THE BIG LIFE COMPANY

Countersignatory Details

Registered Person/Body:
UCHECK

Countersignatory:
ROSY CLARK

Police Records of Convictions, Cautions, Reprimands and Warnings

NONE RECORDED

Information from the list held under Section 142 of the Education Act 2002

NOT REQUESTED

DBS Children's Barred List information

NOT REQUESTED

DBS Adults' Barred List information

NONE RECORDED

Other relevant information disclosed at the Chief Police Officer(s) discretion

NONE RECORDED

Enhanced Certificate

This document is an Enhanced Criminal Record Certificate within the meaning of sections 113B and 116 of the Police Act 1997.

THIS CERTIFICATE IS NOT EVIDENCE OF IDENTITY

Continued on page 2

81892302

Certificate Number 001415330061

Use of certificate information

The information contained in this certificate is confidential and all recipients must keep it secure and protect it from loss or unauthorised access. This Certificate must only be used in accordance with the Disclosure and Barring Service's (DBS) Code of Practice and any other guidance issued by the DBS. Particular attention must be given to the guidance in the fair use of the information in respect of those whose Certificate reveals a conviction or similar information. The DBS will monitor the compliance of Registered Bodies with this Code of Practice and other guidance.

This Certificate is issued in accordance with Part V of the Police Act 1997, which creates a number of offences. These offences include forgery or alteration of Certificates, obtaining Certificates under false pretences, and using a Certificate issued to another person as if it was one's own.

This Certificate is not evidence of identity of the bearer, nor does it establish a person's entitlement to work in the UK.

Certificate content

The personal details contained in this Certificate are those supplied by or on behalf of the person to whom the Certificate relates at the time the application was made and that appear to match any conviction or other details linked to that identity.

The information contained in this Certificate is derived from police records, and from records held of those who are unsuitable to work with children and/or adults, where indicated. The police records are those held on the Police National Computer (PNC) that contains details of Convictions, Cautions, Reprimands and Warnings in England and Wales, and most of the relevant convictions in Scotland and Northern Ireland may also be included. The DBS reserves the right to add new data sources. For the most up to date list of data sources which are searched by the DBS please visit the DBS website.

The Other Relevant Information is disclosed at the discretion of the Chief Police Officers or those of an equivalent level in other policing agencies, who have been approached by the DBS, with due regard to the position sought by the person to whom the Certificate relates.

Certificate accuracy

The DBS is not responsible for the accuracy of police records.

If the person to whom this Certificate relates is aware of any inaccuracy in the information contained in the Certificate, he or she should contact the Countersignatory immediately, in order to prevent an inappropriate decision being made on their suitability. This Countersignatory will advise how to dispute that information, and if requested arrange for it to be referred to the DBS on their behalf. The information should be disputed within 3 months of the date of issue of the Certificate.

The DBS will seek to resolve the matter with the source of the record and the person to whom the Certificate relates. In some circumstances it may only be possible to resolve a dispute using fingerprints, for which consent of the person to whom the Certificate relates will be required.

If the DBS upholds the dispute a new Certificate will be issued free-of-charge. Details of the DBS's disputes and complaints procedure can be found on the DBS's website.

Contact us

Post:	Disclosure and Barring Service PO Box 165 Liverpool L69 3JD	Telephone:	Customer Services: Welsh line: Minicom:	0870 90 90 811 0870 90 90 223 0870 90 90 344
Web:	www.homeoffice.gov.uk/dbs			
Email:	customerservices@dbs.gsi.gov.uk			

If you find this certificate and are not able to return it to the person to whom it relates, please return it to the DBS at the address above or hand it in at the nearest police station.

The information contained in this Certificate may be considered relevant and ought to be considered by those who are unsuitable to work with children and/or adults, where indicated. The police records are those held on the Police National Computer (PNC) that contains details of Convictions, Cautions, Reprimands and Warnings in England and Wales, and most of the relevant convictions in Scotland and Northern Ireland may also be included. The DBS reserves the right to add new data sources. For the most up to date list of data sources which are searched by the DBS please visit the DBS website.

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If the DBS upholds the dispute a new Certificate will be issued free-of-charge. Details of the DBS's disputes and complaints procedure can be found on the DBS's website.

Contact us

Post:	Disclosure and Barring Service PO Box 165 Liverpool L69 3JD	Telephone:	Customer Services: Welsh line: Minicom:	0870 90 90 811 0870 90 90 223 0870 90 90 344
Web:	www.homeoffice.gov.uk/dbs			
Email:	customerservices@dbs.gsi.gov.uk			

If you find this certificate and are not able to return it to the person to whom it relates, please return it to the DBS at the address above or hand it in at the nearest police station.

End of Details

Appendix 5: Recruitment text

1, Facebook status and group use:

"Hi, I'm currently looking for people who have EDS to take part in an interview for my MSc dissertation project. The project is investigating the role of support in managing EDS. If you are interested in taking part, please privately message me and I can send you further information about the study"

2, direct contact - friends

"Hi <name>, wanted to let you know I had posted some information on my dissertation study in my status and on <support group/s> Facebook page. I thought I'd message you just in case you may be interested, but hadn't seen it."

3, face-to-face - support group meeting/s

"Hi, my name is Kate and I'm a postgraduate student at the University of Chester, studying on the MSc Psychology conversion course. I'm currently looking for people who have EDS to take part in an interview looking at the role of support in managing EDS. If you are interested in taking part, please let me know and I can give you more information."

4, poster - see appendix 6.

5, First contact email/ private message

Facebook private message

"Hi <name>, thank you for your interest in this study; I would like to send you an information sheet concerning further information about the research. Please send me details of an email address I can contact you on, or email me at 1521754@chester.ac.uk.

You are under no obligation to take part in the study; should you choose to participate, all identifiable information will be anonymised, and I must stress that the research will have no impact on the services you currently receive or may receive in the future.

If you are interested in participating, we need people to take part in an interview at Altrincham library (alternative text: the University of Chester Parkgate Campus), which could take up to an hour. Let me know if you need information about the location or have any other queries."

Email response to potential participant

"Hi <name>, thank you for your interest in this study; I've attached an information sheet that contains further information about the research. You are under no obligation to take part in the study; should you choose to participate, all identifiable information will be anonymised, and I must stress that the research will have no impact on the services you currently receive or may receive in the future.

If you are interested in participating, we need people to take part in an interview at Altrincham library (alternative text: the University of Chester Parkgate Campus), which could take up to an hour.

Let me know if you need information about the location, have any other queries or would like to be a participant in the study".

7, First follow-up email

"Hi <name>,

I'm following-up on your email of <date>, which expressed interest in my research study into the role of support in managing EDS. Have you had chance to read the participant information sheet I sent you? Do let me know if you need another copy, or did not receive the email with the information sheet attached.

As I mentioned, you are under no obligation to take part in the study; should you choose to participate, all identifiable information will be anonymised, and I must stress that the research will have no impact on the services you currently receive or may receive in the future.

I understand you may not have time to take part in the study, or may not wish to do so; thank you for your interest in the research - please do not hesitate to contact me if you have any queries or concerns about the study, if you need information about the location, have any other queries or would like to be a participant in the study"

8, Second follow-up email (to be sent approx two weeks prior to scheduled end of recruitment and data collection)

"Hi <name>,

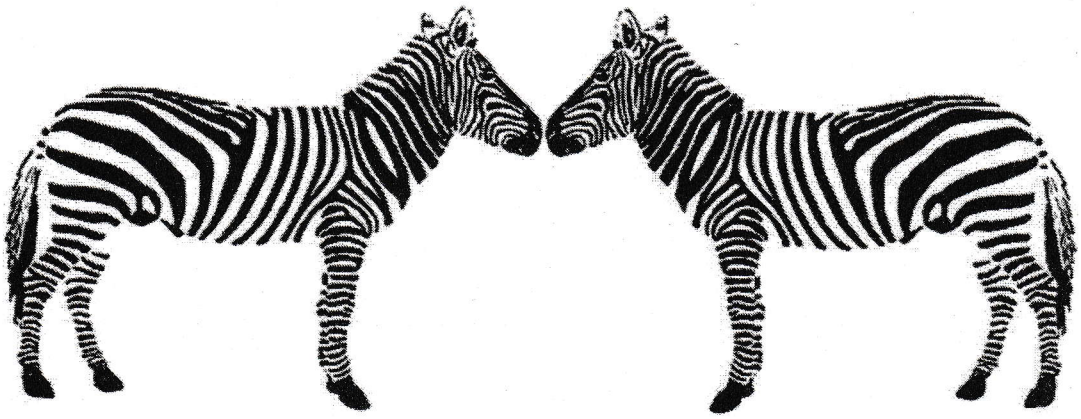
I'm following-up on your email of <date>, and wanted to ask if you were still interested in the research study into the role of support in managing EDS? If you were considering taking part in the study, there is still time to do so as I will be recruiting participants for another two weeks.

As I mentioned, you are under no obligation to take part in the study; should you choose to participate, all identifiable information will be anonymised, and I must stress that the research will have no impact on the services you currently receive or may receive in the future.

I understand you may not have time to take part in the study, or may not wish to do so; thank you for your interest in the research - please do not hesitate to contact me if you have any queries or concerns about the study, whether or not you are considering taking part."

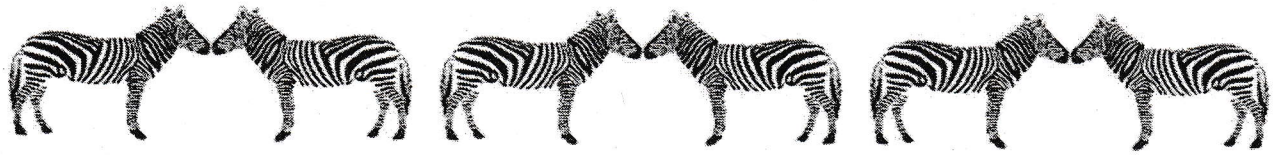
Appendix 6: Recruitment poster

Evaluating the role of external support in helping people with Ehlers-Danlos Syndrome live with their condition



I am currently looking for people with EDS to take part in an interview for a study into how support helps people with EDS live with their condition. This is a dissertation project for MSc Psychology (conversion)

If you are interested in taking part, please email: 1521754@chester.ac.uk for further information



The role of support in managing EDS
Kate Appleby
Email: 1521754@chester.ac.uk

Appendix 7: Consent form

ID:

Evaluating the role of external support in helping people with EDS live with their condition

Consent Form

I understand that the main purpose of this study is to investigate the role of support in helping people with Ehlers-Danlos Syndrome live with their condition. I agree to the following:

- I am over 18
- I consent to the audio recording of the interview
- I understand that I have the right to withdraw from this study until five working days after the interview - by contacting the researcher and quoting my ID code, and that I do not have to give a reason for withdrawing
- I have been informed that my data will be kept confidential, and understand that some comments made may be quoted anonymously
- I have read and understand the information sheet provided and I consent to participate in this study

Print name: _____

Signed: _____ Date: _____

☐ I wish to receive a copy of the results of this study, and consent to the retention of my contact details for this purpose (please tick if valid)

University of Chester

UNIVERSITY OF CHESTER DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL REVIEW FORM

A) Applicant and submission details

Name of applicant: *Kate Appleby*
Project title: *the impact of external support in helping people with EDS live with the condition*

Applicant status: ☐ UG ☒PGT ☐PGR ☐Staff

If you are the applicant's supervisor, have you discussed ethical issues with the applicant?
☐Yes, the applicant is an UG/PGT student and I wish to send the application for accelerated student review.
☐Yes, the applicant is a UG/PGT student and I wish to send the application for full review.
☐Yes, the applicant is a PGR student and I wish to send the application for full review.
☐No → Comments: *Click here to enter text.*

B) Review of application

1. Has the applicant signed and dated the form?

a) ☒Yes → Go to Q2 ☐No → Return to applicant for signature before continuing with review process.

2. What is the submission type?

a) ☒First submission to this or any other committee

b) ☐Resubmission of a rejected application by this committee
↳ Is there a summary of the requirements of the committee? Is the original application attached?: ☐ Yes ☐No→ Return to applicant for full details

c) ☐Revised submission intended to replace an application approved by this committee
↳ Is the original application attached?: ☐ Yes ☐No→ Return to applicant for full details

d) ☐First submission to this committee; has been submitted to another committee.
↳ Is the original application attached? ☐ Yes ☐No→ Return to applicant for full details

3. Research Plan and Methodology

a) Is the study well formulated in terms of drawing on the relevant literature and is it methodologically, analytically and scientifically sound?
☒ Yes ☐No Comments: *Click here to enter text.*

b) Are the timescales provided appropriate?
☒ Yes ☐No Comments: *Click here to enter text.*

c) Are there contingency details?
☒ Yes ☐No Comments: *Click here to enter text.*

d) Is there consideration of how to minimise, manage and monitor issues of distress and harm, however minor?
☒ Yes ☐No Comments: *Click here to enter text.*

e) Are appropriate debrief details provided?

☒ Yes ☐No Comments: *Click here to enter text.*

f) Are appropriate details regarding the use and management of deception provided?

☐ Yes ☐No ☒N/A Comments: *Click here to enter text.*

4. Sample size, participants and recruitment

a) Has the applicant provided appropriate details of the sample and how it will be identified? (this information will be in Q11,Q12, Q13, Q17,Q18, Q20, Q21)

☒ Yes ☐No Comments: *Click here to enter text.*

b) Has the applicant provided appropriate details of where the research will take place, including issues regarding permission and appropriate health and safety information?. Is the necessary documentation attached?

☒ Yes ☐No Comments: *Click here to enter text.*

If the applicant is a taught student and they did not attend the mandatory H&S briefing have they provided appropriate evidence that they have full and satisfactory awareness of the relevant health and safety protocol?

☐ Yes ☐No ☐N/A Comments: *Click here to enter text.*

c) Has the applicant provided appropriate details and attached the necessary documentation concerning their recruitment procedures? In particular, have they appropriately considered how to minimise, manage and monitor issues of distress and harm?

☒ Yes ☐No Comments: *Click here to enter text.*

Are there appropraite RPS credits? ☐ Yes ☐No ☒N/A

d) Has the applicant provided appropriate details and attached the necessary documentation concerning the information made available to participants? In particular, are there appropriate considerations if using internet mediated research?

☒ Yes ☐No Comments: *Click here to enter text.*

Is there appropriate consideration of how to manage issues of distress and harm?

☒ Yes ☐No Comments: *Click here to enter text.*

Are there appropriate details regarding informed consent?

☒ Yes ☐No Comments: *Click here to enter text.*

Are there appropriate details regarding anonymity and confidentiality?

☒ Yes ☐No Comments: *Click here to enter text.*

Are there appropriate details regarding withdrawal procedures?

☒ Yes ☐No Comments: *Click here to enter text.*

e) Are there appropriate details regarding time commitment from participants?

☒ Yes ☐No Comments: *Click here to enter text.*

f) Are there appropriate details regarding compensation arrangements?

☒ Yes ☐No Comments: *Click here to enter text.*

5. Data Collection and Analysis

a) Has the applicant provided full procedural details and attached the necessary documentation concerning data collection procedures?

☒ Yes ☐No Comments: Click here to enter text.

6. Data Analysis

a) Has the applicant provided appropriate details concerning data analysis?

☒ Yes ☐No Comments: *Click here to enter text.*

7. Data protection and Storage

a) Has the applicant provided appropriate details concerning data protection and storage? Have security issues been properly considered?

☒ Yes ☐No Comments: *Click here to enter text.*

8. Dissemination

a) Has the applicant provided appropriate details concerning research dissemination?

☒ Yes ☐No Comments: *Click here to enter text.*

Are there appropriate details regarding how privacy and confidentiality will be maintained during dissemination?

☒ Yes ☐No Comments: *Click here to enter text.*

Are there appropriate details regarding any specific considerations about sharing the research?

☒ Yes ☐No Comments: *Click here to enter text.*

General comments: *A thorough application*

Review status

☐ Chair's action

☐ Staff/PGR for full review ☒ UG/PGT for full review

☐ Work with external agencies ☐ Work with vulnerable participants

☐ Other issues/concerns *Click here to enter text.*

NAME: *Clea Wright*

☐Supervisor ☐Supervisor/Reviewer 1 ☐Reviewer 1 ☒Reviewer 2

DATE: 30/03/2016

University of Chester

UNIVERSITY OF CHESTER DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL REVIEW FORM

A) Applicant and submission details

Name of applicant: *Kate Appleby*
Project title: *The impact of external support in helping people with EDS live with the condition*
Applicant status: ☐ UG ☒PGT ☐PGR ☐Staff
If you are the applicant's supervisor, have you discussed ethical issues with the applicant?
☐Yes, the applicant is an UG/PGT student and I wish to send the application for accelerated student review.
☒Yes, the applicant is a UG/PGT student and I wish to send the application for full review.
☐Yes, the applicant is a PGR student and I wish to send the application for full review.
☐No → Comments: *Click here to enter text.*

B) Review of application

1. Has the applicant signed and dated the form?
a) ☒Yes → Go to Q2 ☐No → Return to applicant for signature before continuing with review process.

2. What is the submission type?
a) ☒First submission to this or any other committee

b) ☐Resubmission of a rejected application by this committee
↳ Is there a summary of the requirements of the committee? Is the original application attached?: ☐ Yes ☐No→ Return to applicant for full details

c) ☐Revised submission intended to replace an application approved by this committee
↳ Is the original application attached?: ☐ Yes ☐No→ Return to applicant for full details

d) ☐First submission to this committee; has been submitted to another committee.
↳ Is the original application attached? ☐ Yes ☐No→ Return to applicant for full details

3. Research Plan and Methodology
a) Is the study well formulated in terms of drawing on the relevant literature and is it methodologically, analytically and scientifically sound?
☒ Yes ☐No Comments: *Click here to enter text.*

b) Are the timescales provided appropriate?
☒ Yes ☐No Comments: *Click here to enter text.*

c) Are there contingency details?
☒ Yes ☐No Comments: *Click here to enter text.*

d) Is there consideration of how to minimise, manage and monitor issues of distress and harm, however minor?
☒ Yes ☐No Comments: *Click here to enter text.*

e) Are appropriate debrief details provided?
☒ Yes ☐No Comments: *Click here to enter text.*

f) Are appropriate details regarding the use and management of deception provided?
☐ Yes ☐No ☒N/A Comments: Click here to enter text.

4. Sample size, participants and recruitment

a) Has the applicant provided appropriate details of the sample and how it will be identified? (this information will be in Q11,Q12, Q13, Q17,Q18, Q20, Q21)
☒ Yes ☐No Comments: Click here to enter text.

b) Has the applicant provided appropriate details of where the research will take place, including issues regarding permission and appropriate health and safety information?. Is the necessary documentation attached?
☒ Yes ☐No Comments: Click here to enter text.
If the applicant is a taught student and they did not attend the mandatory H&S briefing have they provided appropriate evidence that they have full and satisfactory awareness of the relevant health and safety protocol?
☒ Yes ☐No ☐N/A Comments: Click here to enter text.

c) Has the applicant provided appropriate details and attached the necessary documentation concerning their recruitment procedures? In particular, have they appropriately considered how to minimise, manage and monitor issues of distress and harm?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropraite RPS credits? ☐ Yes ☐No ☐N/A

d) Has the applicant provided appropriate details and attached the necessary documentation concerning the information made available to participants? In particular, are there appropriate considerations if using internet mediated research?
☒ Yes ☐No Comments: Click here to enter text.
Click here to enter text.
Is there appropriate consideration of how to manage issues of distress and harm?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropriate details regarding informed consent?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropriate details regarding anonymity and confidentiality?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropriate details regarding withdrawal procedures?
☒ Yes ☐No Comments: Click here to enter text.

e) Are there appropriate details regarding time commitment from participants?
☒ Yes ☐No Comments: Click here to enter text.

f) Are there appropriate details regarding compensation arrangements?
☒ Yes ☐No Comments: Click here to enter text.

5. Data Collection and Analysis

a) Has the applicant provided full procedural details and attached the necessary documentation concerning data collection procedures?
☒ Yes ☐No Comments: Click here to enter text.

6. **Data Analysis**
   a) Has the applicant provided appropriate details concerning data analysis?
   ☒ Yes ☐No Comments: Click here to enter text.

7. **Data protection and Storage**
   a) Has the applicant provided appropriate details concerning data protection and storage? Have security issues been properly considered?
   ☒ Yes ☐No Comments: Click here to enter text.

8. **Dissemination**
   a) Has the applicant provided appropriate details concerning research dissemination?
   ☒ Yes ☐No Comments: Click here to enter text.
   Are there appropriate details regarding how privacy and confidentiality will be maintained during dissemination?
   ☐ Yes ☐No Comments: Click here to enter text.
   Are there appropriate details regarding any specific considerations about sharing the research?
   ☐ Yes ☐No Comments: Click here to enter text.

General comments: It might be worth having posters on campus as part of the contingency plan so think about having a staged approach to recruitment (so maybe put posters up end of April/ beginning of May if recruitment is not going well). If someone privately messages you on Facebook, it is ok to use this to provide them with the link rather than asking them to email your university account. On the consent form, I would ask them to provide their contact details if they would like a summary of the results.

Review status
☐ Chair's action
☐ Staff/PGR for full review ☒ UG/PGT for full review
☐ Work with external agencies ☐ Work with vulnerable participants
☐ Other issues/concerns Click here to enter text.

NAME: Janine Carroll
☒Supervisor ☐Supervisor/Reviewer 1 ☐Reviewer 1 ☐Reviewer 2
DATE: 30/03/2016

University of Chester

UNIVERSITY OF CHESTER DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL REVIEW FORM

A) Applicant and submission details

Name of applicant: *Kate Appleby*
Project title: The impact of external support ...
Applicant status: ☐ UG ☒PGT ☐PGR ☐Staff
If you are the applicant's supervisor, have you discussed ethical issues with the applicant?
☐Yes, the applicant is an UG/PGT student and I wish to send the application for accelerated student review.
☐Yes, the applicant is a UG/PGT student and I wish to send the application for full review.
☐Yes, the applicant is a PGR student and I wish to send the application for full review.
☐No → Comments: *Click here to enter text.*

B) Review of application

1. Has the applicant signed and dated the form?
a) ☒Yes → Go to Q2 ☐No → Return to applicant for signature before continuing with review process.

2. What is the submission type?
a) ☒First submission to this or any other committee

b) ☐Resubmission of a rejected application by this committee
↳ Is there a summary of the requirements of the committee? Is the original application attached?: ☐ Yes ☐No→ Return to applicant for full details

c) ☐Revised submission intended to replace an application approved by this committee
↳ Is the original application attached?: ☐ Yes ☐No→ Return to applicant for full details

d) ☐First submission to this committee; has been submitted to another committee.
↳ Is the original application attached? ☐ Yes ☐No→ Return to applicant for full details

3. Research Plan and Methodology
a) Is the study well formulated in terms of drawing on the relevant literature and is it methodologically, analytically and scientifically sound?
☒ Yes ☐No Comments: *Click here to enter text.*

b) Are the timescales provided appropriate?
☒ Yes ☐No Comments: *Click here to enter text.*

c) Are there contingency details?
☒ Yes ☐No Comments: *Click here to enter text.*

d) Is there consideration of how to minimise, manage and monitor issues of distress and harm, however minor?
☒ Yes ☐No Comments: *Click here to enter text.*

e) Are appropriate debrief details provided?
☒ Yes ☐No Comments: *Click here to enter text.*

f) Are appropriate details regarding the use and management of deception provided?
☐ Yes ☐No ☒N/A Comments: Click here to enter text.

4. Sample size, participants and recruitment

a) Has the applicant provided appropriate details of the sample and how it will be identified? (this information will be in Q11,Q12, Q13, Q17,Q18, Q20, Q21)
☒ Yes ☐No Comments: Click here to enter text.

b) Has the applicant provided appropriate details of where the research will take place, including issues regarding permission and appropriate health and safety information?. Is the necessary documentation attached?
☒ Yes ☐No Comments: Click here to enter text.
If the applicant is a taught student and they did not attend the mandatory H&S briefing have they provided appropriate evidence that they have full and satisfactory awareness of the relevant health and safety protocol?
☐ Yes ☐No ☐N/A Comments: Click here to enter text.

c) Has the applicant provided appropriate details and attached the necessary documentation concerning their recruitment procedures? In particular, have they appropriately considered how to minimise, manage and monitor issues of distress and harm?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropraite RPS credits? ☐ Yes ☐No ☐N/A

d) Has the applicant provided appropriate details and attached the necessary documentation concerning the information made available to participants? In particular, are there appropriate considerations if using internet mediated research?
☒ Yes ☐No Comments: Click here to enter text.

Is there appropriate consideration of how to manage issues of distress and harm?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropriate details regarding informed consent?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropriate details regarding anonymity and confidentiality?
☒ Yes ☐No Comments: Click here to enter text.
Are there appropriate details regarding withdrawal procedures?
☒ Yes ☐No Comments: Click here to enter text.

e) Are there appropriate details regarding time commitment from participants?
☒ Yes ☐No Comments: Click here to enter text.

f) Are there appropriate details regarding compensation arrangements?
☐ Yes ☐No Comments: *It may be a barrier to participation for some that travel expenses are not available. May be worth considering whether interview by phone or Skype might be offered as well? Although recognize that his creates a different interaction. It may be worth considering this – such an addition would require submission of an amendment form*

5. Data Collection and Analysis

a) Has the applicant provided full procedural details and attached the necessary documentation concerning data collection procedures?
☒ Yes ☐No Comments: Click here to enter text.

6. **Data Analysis**
   a) Has the applicant provided appropriate details concerning data analysis?
   ☒ Yes ☐No Comments: *Click here to enter text.*

7. **Data protection and Storage**
   a) Has the applicant provided appropriate details concerning data protection and storage? Have security issues been properly considered?
   ☒ Yes ☐No Comments: *Click here to enter text.*

8. **Dissemination**
   a) Has the applicant provided appropriate details concerning research dissemination?
   ☒ Yes ☐No Comments: *Click here to enter text.*
   Are there appropriate details regarding how privacy and confidentiality will be maintained during dissemination?
   ☒ Yes ☐No Comments: *Click here to enter text.*
   Are there appropriate details regarding any specific considerations about sharing the research?
   ☐ Yes ☐No Comments: *Click here to enter text.*

General comments: *A sensitive project but careful thought has been given. Recommend approval. Applicant will need to submit copies of letters etc granting access.*

Review status
☐ Chair's action
☐ Staff/PGR for full review ☒ UG/PGT for full review
☐ Work with external agencies ☐ Work with vulnerable participants
☐ Other issues/concerns *Click here to enter text.*

NAME: *Ros Bramwell*
☐Supervisor ☐Supervisor/Reviewer 1 ☐Reviewer 1 ☒Reviewer 2
DATE: 05/04/2016

Appendix 3: Ethics Committee Amendment Forms

University of Chester

UNIVERSITY OF CHESTER, DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL AMENDMENT FORM

A) Applicant and personnel

Applicant: *Kate Appleby*
Project title: *The impact of external support in helping people with Ehlers-Danlos Syndrome live with the condition*
Applicant status: ☐ Staff → Go to Section B ☐PGR ☐Undergraduate X Postgraduate taught
Supervisor: *Dr Janine Carroll*

B) Declaration

1. **X** I have submitted an application for ethical approval to the Department of Psychology Ethics Committee and I am required to make the following amendments to my application.
   List the recommendations of the committee. *Chairs Comments: copies of Letters granting access will need to be provided, consider phone/skype interviews, act on and amend with respect to reviewers' comments. Reviewer comment: A thorough application. Supervisor comments: It might be worth having posters on campus as part of the contingency plan, so think about having a staged approach to recruitment. If someone privately messages you on Facebook it is ok to use this to provide them with the link rather than asking them to email your university account. On the consent form, I would ask them to provide their contact details if they would like a summary of the results. Reviewer comment: A sensitive project, but careful thought has been given. Recommend approval. Applicant will need to submit copies of letters etc granting access.*
   Describe how you have addressed these requirements. *Subsequent to my amendment form of 25/05/2016, which addresses the above detailed comments, I have also received permission to recruit from Facebook group: "Manchester Zebras" (please see attached). I will submit additional amendment requests should further permissions be granted.*

2. ☐ I have submitted an application for ethical approval to the Department of Psychology Ethics Committee that was approved on Click here to enter a date.
   I wish the committee to consider the following amendments I would like to make to the research plan (attach the original approved application form) *Click here to enter text.*

☐ I am a member of staff. **Signed:** _____ **Date:** Click here to enter a date.
Print the amendment form on BLUE PAPER and submit to the Dept. Office
X I am an UG/PGT/PGR student. I have discussed any amendments with my project supervisor.
Print the amendment form on BLUE PAPER and submit to the Dept. Office
Signed: Kate Appleby **(Lead Applicant) Date:** 29/05/2016

Supervisor comments:
I have discussed the recommendations of the committee with the applicant and I am satisfied they have met the stated requirements./I support the amendments to the research plan. (delete as appropriate)
☐ Yes Sign and date the form ☐ No Comments: *Click here to enter text.*

I have discussed the recommendations of the committee with the applicant and I am satisfied they have met the stated requirements./I support the amendments to the research plan. (delete as appropriate)

☑ Yes Sign and date the form ☐ No Comments: *Click here to enter text.*

Signed: J. Carroll **(Supervisor)** **Date:** 26/5/16 Click here to enter a date.

COMMITTEE COMMENTS:

☑ **ACCEPTABLE: You may now commence with data collection subject to approval from any relevant external agencies.**

DATA COLLECTION IS NOT PERMISSABLE UNDER THESE CONDITIONS

☐ **ACCEPTABLE SUBJECT TO SUBMISSION OF FURTHER AMENDMENT FORM.**

☐ **Acceptable subject to conditions listed by chair. Discuss conditions highlighted with supervisor and submit ethics application amendment form direct to office.**

☐ **Acceptable subject to conditions listed by chair: Submit ethics application amendment form direct to office.**

Signed: Moira E. Lafferty **Date:** Click here to enter a date. June 2016

ID:

Evaluating the role of external support in helping people with EDS live with their condition

Consent Form

I understand that the main purpose of this study is to investigate the role of support in helping people with Ehlers Danlos Syndrome live with their condition. I agree to the following:

- I am over 18
- I consent to the audio recording of the interview
- I understand that I have the right to withdraw from this study until five working days after the interview - by contacting the researcher and quoting my ID code, and that I do not have to give a reason for withdrawing
- I have been informed that my data will be kept confidential, and understand that some comments made may be quoted anonymously
- I have read and understand the information sheet provided and I consent to participate in this study

Print name: _____

Signed: _____ Date: _____

☐ I wish to receive a copy of the results of this study, and consent to the retention of my contact details for this purpose (please tick if valid, and add contact details below)

Email: _____

Address: _____

_____

_____

Outlook

Search Mail and People

New | Delete Archive Junk | Sweep Move to Categories Undo

Folders

Favourites

Inbox 386
Clutter
Sent Items
Drafts 6

KATE APPLEBY

Inbox 386
Clutter
Drafts 6
Sent Items
Deleted Items
Junk Email 1
Notes

Altrincham library meeting room

GJ Gibbs, Jill <Jill.Gibbs@trafford.gov.uk> |
29/04/2016

Hi Kate

As per our conversation earlier regarding use of the meeting room. I confirm that you will be able to use the meeting room for the purpose of your research.
Once you know the dates please contact me so we can secure the use of the room as we do have regular users.

Regarding the possibility of displaying a poster, once again this is fine

Kind regards

Jill Gibbs
Team Manager
Altrincham/Hale/Partington and Timperley Library

912 5927/5601/5450

Trafford Council is a well-performing, low-cost council delivering excellent services to make Trafford a great place to live, learn, work and relax
You can find out more about us by visiting www.trafford.gov.uk

KA KATE APPLEBY Reply |
29/04/2016
jill.gibbs@trafford.gov.uk <Jill.Gibbs@trafford.gov.uk>

Sent Items

Hi Jill,

I'm a MSc student at the University of Chester, and for my dissertation module I will be conducting a study investigating the role of support in managing Ehlers-Danlos syndrome. We have previously spoken about the possibility of using Altrincham library's meeting room to conduct interviews for the study: as the library is close to public transport links and is wheelchair accessible it is particularly suitable - I am restricted to using locations that are available free-of-charge.

I'm contacting you again to confirm that it is still possible for me to use the meeting room for this purpose?

I intend to conduct these interviews between mid-May to end of July. I anticipate up to 12 interviews, which will require use of the room for up to 2 hours. (The interviews themselves are expected to take up to an hour). I understand that the availability of the room will vary due to regular bookings and other public use of the space.

I would also like to request permission to display a recruitment poster about the study on site, if that is possible?

Do let me know if you would like any further information about the study. Ethical approval for this study has been sought and obtained from the University of Chester, Department of Psychology Ethics Committee, subject to confirmation of provisional permission to use Altrincham library for the interviews, and permission to recruit participants from various groups and locations.

Thank you again for your support: I very much appreciate Altrincham library and Trafford council's commitment to the local community in providing this sort of space for public use.

Many Thanks
Kate Appleby

haz Hay

+ New Message

Shaz Hay is in your contacts

Conversation started 26 April

Kate Smith 26/04/2016 11:30

Hi, I'm currently looking for people who have EDS to take part in an interview for my MSc dissertation project. The project is investigating the role of support in managing EDS. Would it be possible for me to post messages about the study to the group "Hypermobility syndrome- womens only group", to see if any group members are interested in taking part?

If you would like more details (or are interested in taking part yourself) please let me know and I will send you further information about the study.

Many Thanks

Kate Appleby

Shaz Hay accepted your request. 26 April

Shaz Hay 26/04/2016 13:47

If you could send me some more details regarding project and also the post that you are to place in the group, we can then decide if it's appropriate for the group?

Thanks lovely x

Kate Smith 26/04/2016 14:13

Hi Shaz,

I would post: "Hi, I'm currently looking for people who have EDS to take part in an interview for my MSc dissertation project. The project is investigating the role of support in managing EDS. If you are interested in taking part, please privately message me and I can send you further information

Write a reply...

Add Files Add Photos Press Enter to send Reply

English (UK) · Polski · Español · Português (Brasil) · Français (France)

Shaz Hay + New Message

about the study"

Should anyone contact me for further information I would reply with: "Hi <name>, thank you for your interest in this study; I've attached an information sheet that contains further information about the research. You are under no obligation to take part in the study; should you choose to participate, all identifiable information will be anonymised, and I must stress that the research will have no impact on the services you currently receive or may receive in the future.

If you are interested in participating, we need people to take part in an interview at Altrincham library or the University of Chester Parkgate Campus, or alternatively by video messaging. The interview could take up to an hour.

Let me know if you need information about the location, have any other queries, or would like to be a participant in the study".

I have attached the participant information sheet that will be made available to participants and anyone interested in the study - please let me know if you have any concerns I've not addressed.

Thank you very much for considering my request; much appreciated!

Kate

participant-info.docx open - download

27 April

Shaz Hay 27/04/2016 12:49

Hi Kate,
My apologies for the delay!! You can post with "Admin Approved" as long as the indentifiable info is made anonymous, it should be absolutely fine 🙂

Write a reply...

Add Files Add Photos Press Enter to send ☐ Reply

Shaz Hay

+ New Message

made available to participants and anyone interested in the study - please let me know if you have any concerns I've not addressed.

Thank you very much for considering my request; much appreciated!

Kate

participant-info.docx open · download

27 April

Shaz Hay 27/04/2016 12:49

Hi Kate,
My apologies for the delay!! You can post with "Admin Approved" as long as the indentifiable info is made anonymous, it should be absolutely fine 😃
Good luck with your dissertation

27 April

Kate Smith 27/04/2016 19:45

Hi Shaz, thank you very much; so pleased that you are happy for me to post this!

Yes; I will be redacting any identifiable info when I transcribe the interviews - nothing that could identify participants will be reported.

I will post to the group in a couple of weeks, and will post again just before participant recruitment ends.

Thanks again,
Kate

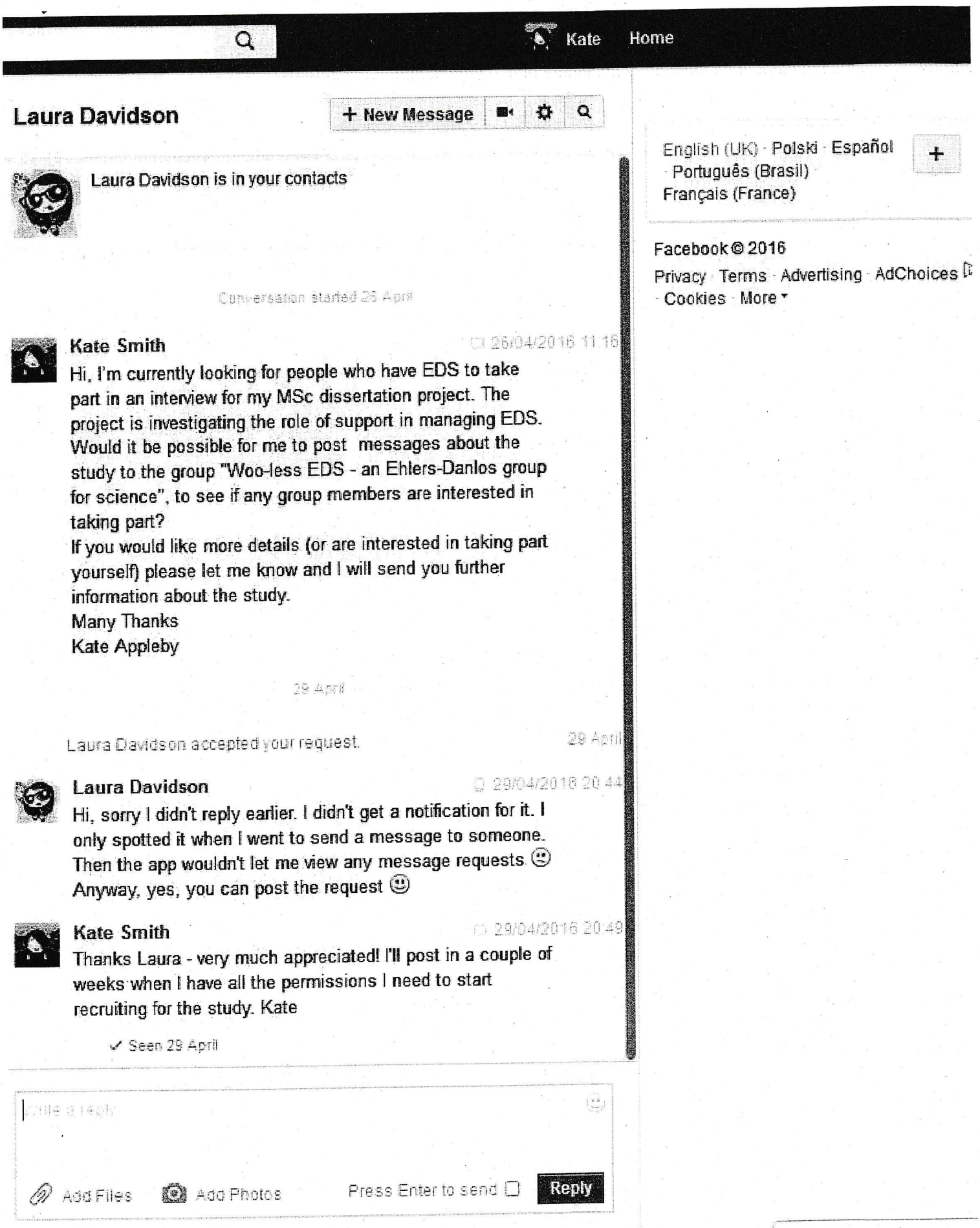
Shaz Hay 27/04/2016 20:01

No worries hun

Sent from Messenger

Add Files Add Photos Press Enter to send Reply

English (UK) · Polski · Español · Português (Brasil) · Français (France)


Kate
Home
Laura Davidson
+ New Message
Laura Davidson is in your contacts
Conversation started 26 April
Kate Smith
26/04/2016 11:16
Hi, I'm currently looking for people who have EDS to take part in an interview for my MSc dissertation project. The project is investigating the role of support in managing EDS. Would it be possible for me to post messages about the study to the group "Woo-less EDS - an Ehlers-Danlos group for science", to see if any group members are interested in taking part?
If you would like more details (or are interested in taking part yourself) please let me know and I will send you further information about the study.
Many Thanks
Kate Appleby
29 April
Laura Davidson accepted your request.
29 April
Laura Davidson
29/04/2016 20:44
Hi, sorry I didn't reply earlier. I didn't get a notification for it. I only spotted it when I went to send a message to someone. Then the app wouldn't let me view any message requests.
Anyway, yes, you can post the request
Kate Smith
29/04/2016 20:49
Thanks Laura - very much appreciated! I'll post in a couple of weeks when I have all the permissions I need to start recruiting for the study. Kate
Seen 29 April
Add Files
Add Photos
Press Enter to send
Reply
English (UK) · Polski · Español · Português (Brasil) · Français (France)
Facebook © 2016
Privacy · Terms · Advertising · AdChoices · Cookies · More

University of Chester

UNIVERSITY OF CHESTER, DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL AMENDMENT FORM

A) Applicant and personnel

Applicant: *Kate Appleby*
Project title: *The impact of external support in helping people with Ehlers-Danlos Syndrome live with the condition*
Applicant status: ☐ Staff → Go to Section B ☐PGR ☐Undergraduate X Postgraduate taught
Supervisor: *Dr Janine Carroll*

B) Declaration

1. **X** I have submitted an application for ethical approval to the Department of Psychology Ethics Committee and I am required to make the following amendments to my application.
   List the recommendations of the committee. *Chairs Comments: copies of Letters granting access will need to be provided, consider phone/skype interviews, act on and amend with respect to reviewers' comments. Reviewer comment: A thorough application. Supervisor comments: It might be worth having posters on campus as part of the contingency plan, so think about having a staged approach to recruitment. If someone privately messages you on Facebook it is ok to use this to provide them with the link rather than asking them to email your university account. On the consent form, I would ask them to provide their contact details if they would like a summary of the results. Reviewer comment: A sensitive project, but careful thought has been given. Recommend approval. Applicant will need to submit copies of letters etc granting access.*
   Describe how you have addressed these requirements. *Subsequent to my amendment form of 25/05/2016, which addresses the above detailed comments, I have also received permission to recruit from Facebook group: "Manchester Zebras" (please see attached). I will submit additional amendment requests should further permissions be granted.*

2. ☐ I have submitted an application for ethical approval to the Department of Psychology Ethics Committee that was approved on Click here to enter a date.
   I wish the committee to consider the following amendments I would like to make to the research plan (attach the original approved application form) *Click here to enter text.*

☐ I am a member of staff. **Signed:** _____ **Date:** Click here to enter a date.
Print the amendment form on BLUE PAPER and submit to the Dept. Office
X I am an UG/PGT/PGR student. I have discussed any amendments with my project supervisor.
Print the amendment form on BLUE PAPER and submit to the Dept. Office
Signed: Kate Appleby **(Lead Applicant) Date:** 29/05/2016

Supervisor comments:
I have discussed the recommendations of the committee with the applicant and I am satisfied they have met the stated requirements./I support the amendments to the research plan. (delete as appropriate)
☐ Yes Sign and date the form ☐ No Comments: *Click here to enter text.*

Signed: J. Carroll (Supervisor) Date: 2/6/16 Click here to enter a date.

COMMITTEE COMMENTS:

☑ **ACCEPTABLE: You may now commence with data collection subject to approval from any relevant external agencies.**

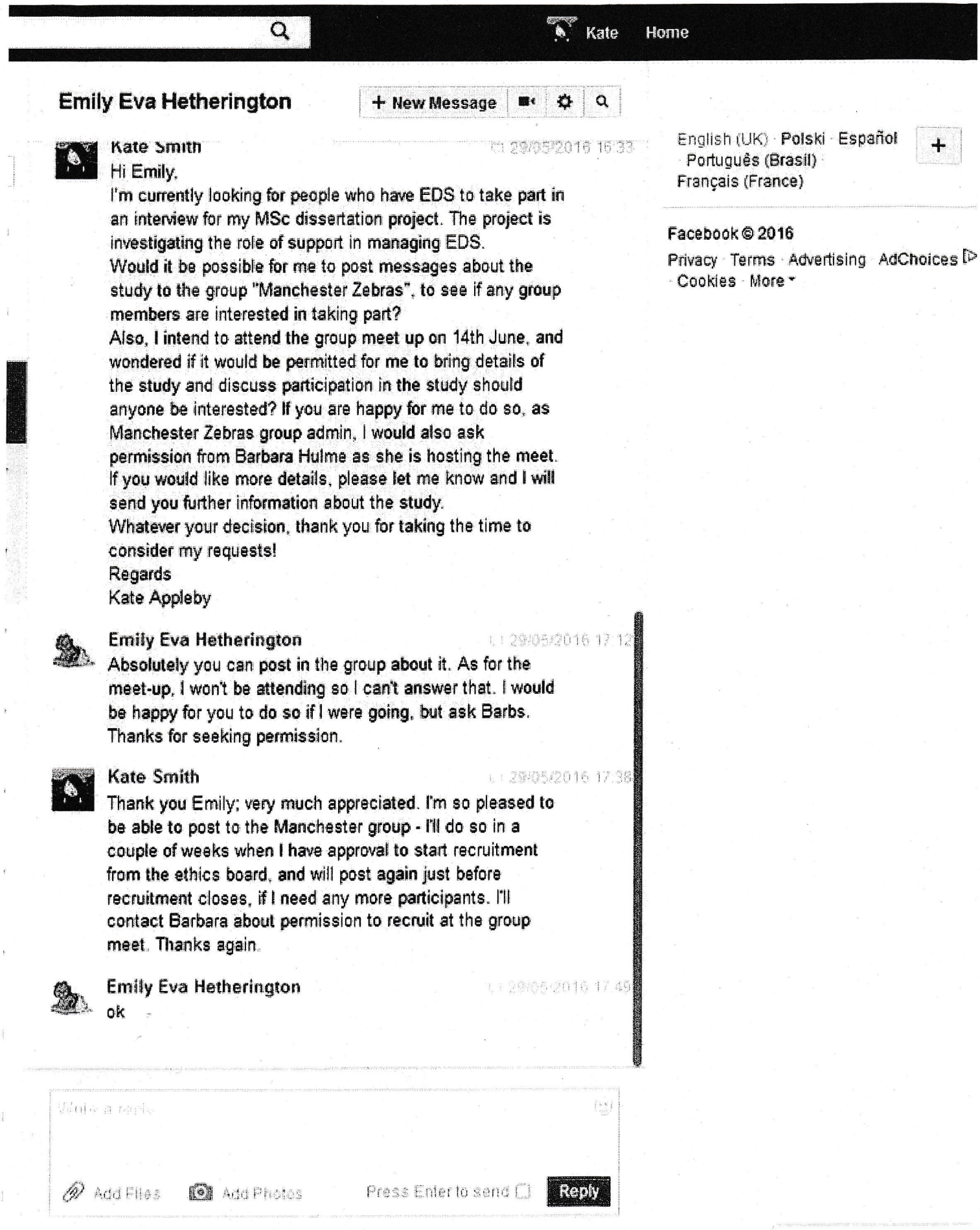
DATA COLLECTION IS NOT PERMISSABLE UNDER THESE CONDITIONS

☐ **ACCEPTABLE SUBJECT TO SUBMISSION OF FURTHER AMENDMENT FORM.**

☐ **Acceptable subject to conditions listed by chair. Discuss conditions highlighted with supervisor and submit ethics application amendment form direct to office.**

☐ **Acceptable subject to conditions listed by chair: Submit ethics application amendment form direct to office.**

Signed: Moira E. Lafferty **Date:** Click here to enter a date. 3/6/14


Kate
Home
Emily Eva Hetherington
+ New Message
Kate Smith
29/05/2016 16:33
Hi Emily,
I'm currently looking for people who have EDS to take part in an interview for my MSc dissertation project. The project is investigating the role of support in managing EDS.
Would it be possible for me to post messages about the study to the group "Manchester Zebras", to see if any group members are interested in taking part?
Also, I intend to attend the group meet up on 14th June, and wondered if it would be permitted for me to bring details of the study and discuss participation in the study should anyone be interested? If you are happy for me to do so, as Manchester Zebras group admin, I would also ask permission from Barbara Hulme as she is hosting the meet.
If you would like more details, please let me know and I will send you further information about the study.
Whatever your decision, thank you for taking the time to consider my requests!
Regards
Kate Appleby
Emily Eva Hetherington
29/05/2016 17:12
Absolutely you can post in the group about it. As for the meet-up, I won't be attending so I can't answer that. I would be happy for you to do so if I were going, but ask Barbs.
Thanks for seeking permission.
Kate Smith
29/05/2016 17:36
Thank you Emily; very much appreciated. I'm so pleased to be able to post to the Manchester group - I'll do so in a couple of weeks when I have approval to start recruitment from the ethics board, and will post again just before recruitment closes, if I need any more participants. I'll contact Barbara about permission to recruit at the group meet. Thanks again.
Emily Eva Hetherington
29/05/2016 17:49
ok
Write a reply...
Add Files
Add Photos
Press Enter to send
Reply
English (UK) · Polski · Español · Português (Brasil) · Français (France)
Facebook © 2016
Privacy · Terms · Advertising · AdChoices · Cookies · More

University of Chester

UNIVERSITY OF CHESTER, DEPARTMENT OF PSYCHOLOGY
APPLICATION FOR ETHICAL APPROVAL AMENDMENT FORM

A) Applicant and personnel

Applicant: *Kate Appleby*
Project title: *The impact of external support in helping people with EDS live with the condition*
Applicant status: ☐ Staff → Go to Section B ☐PGR ☐Undergraduate ☒Postgraduate taught
Supervisor: *Dr Janine Carroll*

B) Declaration

1. ☐ I have submitted an application for ethical approval to the Department of Psychology Ethics Committee and I am required to make the following amendments to my application.
   List the recommendations of the committee. *Click here to enter text.*
   Describe how you have addressed these requirements. *Click here to enter text.*

2. ☒ I have submitted an application for ethical approval to the Department of Psychology Ethics Committee that was approved on 01/06/2016
   I wish the committee to consider the following amendments I would like to make to the research plan (attach the original approved application form) *I am conducting interviews at two specified locations some of which are not convenient for all participants therefore I wish to do interviews at a convenient location for my participants*

☐ I am a member of staff. **Signed:** _____ **Date:** Click here to enter a date.
Print the amendment form on BLUE PAPER and submit to the Dept. Office
☒ I am an UG/PGT/PGR student. I have discussed any amendments with my project supervisor.
Print the amendment form on BLUE PAPER and submit to the Dept. Office

Signed: Kate Appleby **(Lead Applicant) Date:** 15/06/2016

Supervisor comments:
I have discussed the recommendations of the committee with the applicant and I am satisfied they have met the stated requirements./I support the amendments to the research plan. (delete as appropriate)
☒ Yes Sign and date the form ☐ No Comments: *Click here to enter text.*

Signed: J Carroll **(Supervisor) Date:** 15/06/2016

COMMITTEE COMMENTS:

☐ **ACCEPTABLE: You may now commence with data collection subject to approval from any relevant external agencies.**

DATA COLLECTION IS NOT PERMISSABLE UNDER THESE CONDITIONS

☐ **ACCEPTABLE SUBJECT TO SUBMISSION OF FURTHER AMENDMENT FORM.**

☐ **Acceptable subject to conditions listed by chair. Discuss conditions highlighted with supervisor and submit ethics application amendment form direct to office.**

☐ **Acceptable subject to conditions listed by chair: Submit ethics application amendment form direct to office.**

Signed: Moira Flaherty **Date:** Click here to enter a date. 16/6/16

Appendix 4: Participant Information Sheet and Consent form

Evaluating the role of external support in helping people with EDS live with their condition

Participant Information Sheet

You are being invited to participate in a research study. Before you decide whether to participate, it is important for you to understand why the research is being done and what it will involve. Please take the time to read the following information carefully and feel free to ask the interviewer if you would like any further information, or if there is anything you do not understand. I would like to stress that you do not have to accept this invitation, and should only take part if you want to.

Thank you for reading this.

What is the purpose of this study?

This study is evaluating how support can help people with Ehlers Danlos Syndrome (EDS), manage to live with their condition, and what your experiences of different types of support have been.

What will I be asked to do?

After having read this information sheet you will be asked to sign a consent form, and then we will begin the interview. If you have not participated in a research study of this kind before, this interview may seem quite different to other interviews you may have had in the past; they are not diagnostic tools, therapy, or for developmental purposes, and there is no set time for your answers so you can answer in as much detail as you wish to. The interviews are expected to last up to an hour. The questions are about what you understand by the term 'support', and how you have experienced support. The interview will be recorded, and transcribed for use in this study. Your name and any potentially identifying information (such as location or the names of people you may talk about) will be removed from the transcript, so you will not be identifiable as a study participant. When the interview is complete you will be asked to read an information sheet containing further information.

Who is conducting this study?

As part of the dissertation module, Kate Appleby, a postgraduate student on the MSc Psychology (Conversion) course at the University of Chester will be conducting and transcribing the interviews.

Is participation voluntary?

Participation in the study is entirely voluntary and you will not receive any formal compensation or reward (financial or otherwise) for your time. You can withdraw from the study at any time before, or during, the interview and any data that has been collected prior to your withdrawal will be destroyed. You do not have to provide a reason for withdrawing, and you can do so by speaking to the researcher during the interview, or alternatively by email. If you wish to withdraw after the interview, email Kate Appleby (1521754@chester.ac.uk), or my supervisor Dr Janine Carroll (email:

j.carroll@chester.ac.uk) quoting your unique ID. You will have the right to withdraw from this study until five working days after completion of your interview; after this time, the interview will be transcribed and you will not be able to withdraw as the interview will be part of the study analysis.

If for any reason you do not wish to complete the interview, or do not wish to answer a question, but do not wish to withdraw from the study, you can say so at any time during the interview. You will be asked to confirm that you consent to the retention of the interview recording and then to read an information sheet containing further information.

What are the risks/ benefits of taking part in the study?

Many people find taking part in research studies to be a satisfying experience and I hope this will be the same for you.

The interview will be about your experiences of support subsequent to your diagnosis of EDS; ***the researcher will not ask you about how EDS, or the diagnosis of EDS, has impacted your life***. This interview is not expected to cause any harm or distress, however ***if you find talking about EDS upsetting, or if you have experienced distressing or stressful evaluations (e.g. ATOS or healthcare professionals), or think talking about support may upset you, please do not take part in this study.***

Who can take part?

Any adult (18+) who has been diagnosed with any variant of Ehlers Danlos Syndrome (EDS), for over 12 months, may take part in this study.

What about the results?

The information collected will be analysed and although some comments made may be quoted in subsequent scientific publications, this will be done anonymously. Only myself as the researcher, my supervisor (Dr Janine Carroll, email: j.carroll@chester.ac.uk), and other academic staff such as an external examiner will have access to the transcript and audio recordings. If you would like to know more about the results of the study, you can give permission for us to retain your contact details for the purpose of sending you a copy. The results will be written in the form of a one page summary of what has been found.

What about confidentiality?

All interview recordings and your personal details are confidential, and will be kept secure by the researcher in password protected devices.

What if I have further questions?

If you have further questions, or any concerns about this study, please feel free to contact Kate Appleby (email: 1521754@chester.ac.uk) who will try to answer any queries you may have, alternatively you may contact my supervisor, Dr Janine Carroll (email: j.carroll@chester.ac.uk).

Ethical Approval

Ethical approval for this study has been sought and obtained from the University of Chester, Department of Psychology Ethics Committee.

ID:

Evaluating the role of external support in helping people with EDS live with their condition

Consent Form

I understand that the main purpose of this study is to investigate the role of support in helping people with Ehlers Danlos Syndrome live with their condition. I agree to the following:

- I am over 18
- I consent to the audio recording of the interview
- I understand that I have the right to withdraw from this study until five working days after the interview – by contacting the researcher and quoting my ID code, and that I do not have to give a reason for withdrawing
- I have been informed that my data will be kept confidential, and understand that some comments made may be quoted anonymously
- I have read and understand the information sheet provided and I consent to participate in this study

Print name: _____

Signed: _____ Date: _____

☐ I wish to receive a copy of the results of this study, and consent to the retention of my contact details for this purpose (please tick if valid, and add contact details below)

Email: _____

Address: _____

_____

_____

ID:

Appendix 5: Debrief package

Evaluating the role of external support in helping people with EDS live with their condition

Further Information for Participants

Thank you for participating in this research study. The interview is now finished, before we conclude the session, please take the time to read the following information carefully and feel free to ask the researcher if you have any further questions, or have any concerns.

What is the purpose of this study?

This study is evaluating how support can help people with Ehlers-Danlos Syndrome (EDS) manage their condition, and what participant's experiences of different types of support have been.

Many people find taking part in this kind of research to be a satisfying experience and I hope this has been your experience. Basic research of this type can be useful for the provision of evidence based care, and is essential for demonstrating the need for further research into, and specialised services for, people with EDS and other complex chronic conditions.

if you have any further questions or concerns about this study, or its purpose, please contact myself (Kate Appleby, email: 1521754@chester.ac.uk) or my supervisor (Dr. Janine Carroll, email: j.carroll@chester.ac.uk).

What happens next?

The audio recording of the interview will be transcribed, and any details that could make it possible to identify you will be removed; although some comments made may be quoted in subsequent scientific publications, this will be done anonymously. The transcript will be analysed, along with the transcripts of interviews with other participants, and the results will be written up and reported.

At the start of this session you were asked to sign a consent form, which asked if you would like to receive a one page summary of the findings from this study; if you would like to change your answer to that question, let the interviewer know now, as you will not be able to amend this choice once the session has finished.

What if I decided to withdraw from the study?

Participation in the study is entirely voluntary, and you can withdraw from the study at any time before or during the interview, and up to five working days afterwards. *You do not have to provide a reason for withdrawing*, and you may have done so by speaking to the researcher during the interview. If you wish to withdraw after the interview has finished, email Kate Appleby (1521754@chester.ac.uk), or my supervisor Dr Janine Carroll (email: j.carroll@chester.ac.uk) quoting your unique ID. After five working days the interview will be transcribed and you will not be able to withdraw as the interview will be part of the study analysis. If you decide to withdraw, any data recorded will be destroyed.

What if I have concerns about issues raised during the interview, such as the support available to me, or if I feel upset?

This interview is not expected to have caused harm or distress, however if you have found it to be upsetting, or troubling in any way, there are several avenues of support listed below:

Support for EDS:

Ehlers-Danlos Support UK: website: www.ehlers-danlos.org; helpline: 0800 907 8518 Monday, Tuesday, Wednesday and Friday (09:30 – 17:30)

HMSA (Hypermobility Syndromes Association): website: http://hypermobility.org; helpline: 03330 116 388

Support for mild to moderate Anxiety and Depression, or problems with sleep:

CBT programmes, such as 'Beating the Blues' or 'Sleepio' are free to access via the NHS; should you wish to access these short courses, ask your GP about how that can be arranged in your region. Many areas also offer other services such as crisis support, peer support, drop-in groups, and follow-up services for people who have previously accessed such courses.

In many areas of the North West of England these services can also be accessed via: **Self Help Services (part of the Big Life Group)**: website: www.selfhelpservices.org.uk; helpline: 0300 003 7029

Other support:

Samaritans are available 24 hours a day, 365 days a year –*"Just having someone to talk to that isn't family or friends can be a tremendous help. You do not need to be suicidal to get in touch."* Website: www.samaritans.org; helpline: 116 123

You can also contact your GP, or consultant, if you have concerns about any issues raised during the interview.

What if I have further questions?

If you have further questions, or any concerns about this study, please feel free to contact Kate Appleby (email: 1521754@chester.ac.uk) who will try to answer any queries you may have, alternatively you may contact my supervisor, Dr Janine Carroll (email: j.carroll@chester.ac.uk).

Thank you for participating in this study